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Investigating Parenting Stress And Neurodevelopment In Infants With Congenital Heart Defects During The First Year Of Life

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Abstract
Background: Parents of infants with CHD, the most prevalent group of congenital anomalies, experience increased parenting stress levels. These can potentially interfere with the normal parenting process, and with establishing a healthy parent-child relationship, which are important for proper development during infancy. Neurodevelopmental delays are among the major morbidities of children with CHD. The changes in stress over the critical period of infancy (first year of life), and how the stress affects infant development, however, have yet to be studied. Aims: To describe and compare parenting stress levels and changes over time between parents of infants with CHD and parents of healthy infants, during the first year of infants' life; and to examine associations between parenting stress and infant neurodevelopmental outcomes in these populations. Methods: A secondary analysis of data a larger prospective cohort study (N=241), performed during 2003-2007 at the Children's Hospital of Philadelphia, included mixed-effects and general linear regression modeling. Findings: Parents of infants with CHD had higher parenting stress than parents of healthy infants on the Child and Parent Domains at three months of age. The stress remained higher on the Demandingness subscale throughout the first year of infants' life. The change in parenting stress over time significantly differed in parents of infants with CHD and in parents of healthy infants on the Child and Parent Domains, and on the Life Stress. Parents of CHD infants demonstrated decrease in stress over time, and parents of healthy infants generally experienced increase in stress with time. As for the associations between stress and development, findings demonstrate cross-sectional relationships between stress and development, as well as temporal relationships between early stress and later development in both subjects and controls. Conclusions: Findings highlight stressful periods, which may be risky for parents of infants with CHD, and introduce psychosocial/familial aspects as additional contributors to infant development. Family systems intervention may promote parental adaptive coping and productive parenting practices in this population.

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INVESTIGATING PARENTING STRESS AND NEURODEVELOPMENT IN INFANTS WITH CONGENITAL HEART DEFECTS DURING THE FIRST YEAR OF LIFE

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ABSTRACT

INVESTIGATING PARENTING STRESS AND NEURODEVELOPMENT IN INFANTS WITH CONGENITAL HEART DEFECTS DURING THE FIRST YEAR OF LIFE

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Background: Parents of infants with CHD, the most prevalent group of congenital anomalies, experience increased parenting stress levels. These can potentially interfere with the normal parenting process, and with establishing a healthy parent-child relationship, which are important for proper development during infancy. Neurodevelopmental delays are among the major morbidities of children with CHD. The changes in stress over the critical period of infancy (first year of life), and how the stress affects infant development, however, have yet to be studied. **Aims:** To describe and compare parenting stress levels and changes over time between parents of infants with CHD and parents of healthy infants, during the first year of infants' life; and to examine associations between parenting stress and infant neurodevelopmental outcomes in these populations. **Methods**: A secondary analysis of data a larger prospective cohort study (N=241), performed during 2003-2007 at the Children's Hospital of Philadelphia, included mixed-effects and general linear regression modeling. **Findings**: Parents of infants with CHD had higher parenting stress than parents of healthy infants on the Child and Parent Domains at three months of age. The stress remained higher on the Demandingness subscale throughout the first year of infants' life. The change in

parenting stress over time significantly differed in parents of infants with CHD and in parents of healthy infants on the Child and Parent Domains, and on the Life Stress. Parents of CHD infants demonstrated decrease in stress over time, and parents of healthy infants generally experienced increase in stress with time. As for the associations between stress and development, findings demonstrate cross-sectional relationships between stress and development, as well as temporal relationships between early stress and later development in both subjects and controls. **Conclusions**: Findings highlight stressful periods, which may be risky for parents of infants with CHD, and introduce psychosocial/familial aspects as additional contributors to infant development. Family systems intervention may promote parental adaptive coping and productive parenting practices in this population.

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CHAPTER 1: INTRODUCTION TO THE PROBLEM

Background

Although frequently perceived as joyful and satisfying, the parenting process is stressful at times, involving feelings of hopelessness or frustration (Lawoko & Soares, 2002). Parenting stress is defined as the psychological distress one experiences, while trying to meet the demands of the parenting role (Deater-Deckard, 2004). Parenting stress is distinct from other kinds of situational stress, and its effects can be measured separately from other global assessments of stressful circumstances (Deater-Deckard, 1998). Although most parents experience parenting stress to some degree as part of the normal parenting process (Eronen, Pincombe, Calabretto, 2007), parenting stress tends to increase under certain situations such as severe pediatric illness (Deater-Deckard, 2004), thus putting parents, children, and other family members at risk for adverse outcomes.

Parents of children with severe or chronic health conditions (e.g. mental health problems, congenital syndromes, cancer, diabetes, etc.) reported increased levels of parenting stress across studies (Chiou & Hsieh, 2008; Maas-van, Schaaijk, Roeleveld-Versteegh &Van Baar, 2013; Rabineau, Mabe &Vega, 2008; Rimmerman & Stanger, 2001; Torowicz, Irving, Hanlon, Sumpter, Medoff-Cooper, 2010). Those families often face frequent medical interventions, hospitalizations, or special care needs, thus forcing them to deal with the financial, social, and emotional challenges the illness presents (Fonseca, Nazaré, Canavarro, 2011; U.S. Department of Health and Human Services, 2013). Their parenting stress frequently results from illness-related burden and increased care demands.

Increased parenting stress was associated with decreased physical and psychological well-being, often predicting anxiety and depression in both children and parents (Deater-Deckard, 1998; Fonseca et al, 2011). High levels of parenting stress were also predictive of low social competence and maladaptive behaviors among children (Cabrera & Mitchell, 2009; Hintermair, 2006; Semke, Garbacz, Kwon, Sheridan & Woods, 2010). Recent research discovered that parenting stress negatively affects children's development (Grunau, Whitfield, Petrie-Thomas, 2009; Guajardo, Snyder & Petersen, 2009; Molfese, et al., 2010). The common perception is that increased parenting stress drives parents to use maladaptive parenting practices (Abidin, 1992; Farmer & Lee, 2011), which in turn, might disturb the establishment of a healthy parent-child relationship (Cohen, et al, 1999). Thus, decreasing parenting stress in order to support a healthy parent-child relationship early in life is essential for establishing proper development (Bowlby, 1988; Erikson, 1963; Piaget, 1954).

With recent improvements in technology and healthcare, more infants with once fatal conditions are surviving into childhood (Cohen et al., 2011). Only within two decades, the survival rate among infants with congenital heart defects (CHD), the most prevalent group of congenital anomalies occurring in approximately one percent of live births, has increased dramatically from 15% to more than 85% (Van der Bom et al., 2010). The quality-of-life issues and the developmental implications these families face, can be long lasting. The literature reveals significantly higher levels of parenting stress among parents of infants and children with CHD, compared to parents of healthy controls (Mullen et al., 2014; Sarajuuri et al, 2012; Uzark & Jones, 2003). The change in this stress over the

critical period of infancy (first year of life), and how the stress affects infant development, however, have yet to be studied. The proposed study aims to examine parenting stress change over time in parents of infants with CHD, and the relations of parenting stress to infants' neurodevelopment, over the first year of life.

Statement of the Problem

Whether diagnosed prenatally or shortly after the infant is born, CHD carries profound meanings to families, and is often referred to as a "shock' or as a "burden" (Farley et al, 2007; Hayes & Knox, 1984). Even if the infant is prenatally diagnosed, parents may still grieve over the lost dream of a healthy baby, feel guilty or ashamed (Brosig, Whitstone, Frommelt, Frisbee & Leuthner, 2007; Upham, & Medoff-Cooper, 2005). Although diagnoses vary in category and severity, many of these infants undergo complex surgical procedures and long-term hospitalizations in the intensive care environment within their first year of life. Parents of infants with CHD are faced with multiple stressors, including the infants' post-operative fragile condition, intimidating appearance, long separation in the intensive care environment, increased care demands post-discharge, feeding difficulties, and often irritable temperaments (Upham, & Medoff-Cooper, 2005; Lobo, 1992; Torowicz, et al. 2010). These factors may have implications on the relationships being formed between the parents and the infant, by negatively affecting the bonding process (Kennell & Klaus, 1984). Studies investigating family relations within the context of a chronic or life-threatening illness reported strains in the parental role (Miles, Carter, Hennessey, Eberly & Riddle, 1989; Goldberg, Morris, Simmons, Fowler, Levison, 1990). Specifically, the quality of the parent-child relationship

and the parental practices are negatively influenced by parental maladjustment to their child's CHD (Apley, Barbour, & Westmacott, 1967). Developmental theoreticians and researchers agree that disturbed parent-child relationship and child-rearing malpractices can negatively impact child's development (Bowlby, 1988; Darling & Steinberg, 1993; Erikson, 1963; Masten & Coatsworth, 1998; Rapee, 1997).

Children with CHD who have undergone cardiac surgery as infants display a wide range of cognitive, psychosocial, and behavioral abnormalities. These developmental delays (referred to as not meeting developmental milestones, standardized to the general population) include motor and speech delays, executive function deficits, inattention, and hyperactivity. As they get older, these children suffer from learning disabilities and low academic achievement, and frequently need special services in school (Brown, Wernovsky, Mussatto, & Berger, 2005; Wernovsky, 2006; Ballweg, Wernovsky & Gaynor 2007). When combined, these developmental delays represent the most common morbidity in school-aged children with CHD (more common than late mortality, bacterial endocarditis, or arrhythmias) (Wernovsky, 2006). Whereas research has shown that parenting stress is among the predictors of such adverse developmental outcomes in other pediatric populations with health conditions (Grunau et al., 2009; Hintermair, 2006; Molfese, et al., 2010; Voigt, Brandl, Pietz, Pauen, Kliegel, & Reuner, 2013), such associations have yet to be explored in infants with CHD.

Parenting stress in CHD pediatric populations was scarcely investigated as compared to populations with other health conditions (e.g. Autism Spectrum Disorders; Low Birth Weight infants). The existing research in CHD pediatric populations mostly

focuses on the parents, by exploring how parental practices (Rimmerman & Stanger, 2001) and illness-related coping mechanisms (Mullen et al, 2014; Phipps & Drotar, 1990) affect parenting stress. Studies that sought sources leading to increased stress levels found them to be related to the illness' diagnostic characteristics (Sarajuuri, et al. 2012); caretaking burden (Smith, Hefley & Anand, 2007); temperament and other child's characteristics (Torowicz, et al. 2010); parental and familial factors (Dudek-Shriber, 2004); and social contexts (Visconti, et al., 2002). Nonetheless, studies investigating outcomes of increased parenting stress are lacking in the population of CHD children. Moreover, many cross-sectional studies reported limited ability in drawing meaningful conclusions regarding what happens to stress over time (Uzark & Jones, 2003). These studies recommended longitudinal research in the future in order to better understand what happens to parenting stress over time, while accounting for diagnostic features and child's age (Uzark & Jones, 2003).

Purpose of the Study

The purpose of this study is to examine parenting stress in parents of infants with CHD; and the associations between parenting stress and infants' neurodevelopment over the infants' first year of life, through a secondary analysis of data from a larger prospective cohort study. The parent study was conducted during 2003-2007 in the Cardiac Center of the Children's Hospital of Philadelphia, and originally aimed to investigate feeding behaviors and neurodevelopment of infants with CHD. Data were collected via clinical assessments of the infants' diagnostic parameters, anthropometrics, neurodevelopment (mental and psycho-motor development), and feeding parameters. Parents filled out

standardized questionnaires in order to assess parenting stress. Mixed-Effects and General Regression Modeling was performed in the current to detect change in stress over time, and significant associations between stress and development.

Specific Aims

1. Describe and compare parenting stress between parents of infants with CHD and parents of healthy controls at three, six, nine, and twelve months of infants' life.
2. Identify changes in parenting stress in parents of infants with CHD over the first year of infants' life, and compare them to those of healthy controls.
3. Examine associations between parenting stress (at three, six, nine, and twelve months) and infant neurodevelopment (at six and twelve months), in CHD and healthy infants.

Significance

Parenting stress has gained a major research interest in recent years, given the significant impact it has on parents, children and other family members. The dramatic increase in CHD survival rates over the last two decades adds chronic characteristics to the most prevalent congenital anomaly, making the long-term implications of parenting stress more relevant than ever. Though the fact that parents of children with CHD experience increased levels of parenting stress is recognized, we do not know what happens to the stress over time, given the lack of longitudinal assessments in the sensitive period of infancy. Moreover, although parenting stress was demonstrated to have multiple adverse

outcomes on other pediatric illnesses groups, these potential outcomes were not explored in the growing population of children with CHD. Although the study utilized a secondary data analysis, and therefore possesses several design and sampling limitations, it has a merit in addressing gaps in the literature. To my knowledge, the current study was the first to examine the effect of parenting stress on neurodevelopmental outcomes in infants with CHD.

The current study may expand our understanding regarding the stress experienced by parents over time following the sensitive post-diagnostic/surgical period. Identifying the most stressful periods for parents is relevant for everyone treating or coping with CHD. Healthcare providers should consider these potentially increased times of stress for parents whenever administrating clinical protocols, providing information, educating or consenting parents. Parenting stress assessments could be directed specifically to such periods, in order to provide effective anticipatory guidance; stress-reduction interventions to parents; involve other professionals (i.e., therapists, social workers); and support other family members as risk.

Furthermore, identification of the effect of parenting stress on neurodevelopment of infants with CHD can illuminate additional factors contributing to the developmental delays in this group. While the relationship between parental factors and child outcomes has been established in other population, they have not been done so for infants with CHD. In addition, developmental delays are mostly attributed to biology and illness-related clinical parameters. Introducing parenting stress as a factor may convince stakeholders to incorporate social/familial aspect to policies directed to deal with

developmental delays in this growing population. As for nursing clinical practice, once critical periods of peak stress for parents of CHD infant are identified, developmental assessments and nursing stress-relief interventions may be timely directed for greatest efficacy.

CHAPTER 2: BACKGROUND LITERATURE

Conceptual Framework

The Parenting Stress Model

The most familiar model of parenting stress was developed by Abidin in 1976 (see figure 1). Abidin identified specific stress-evoking factors in the parenting role, and categorized them into several domains. These domains are related to the parents, to the child, and to the situation (or general life events) (Abidin, 1995). Stressors in the child domain involve temperamental and behavioral factors of the child, as well as parents' perceptions and expectations with regard to their child and their parental role. These factors include the child's adaptability (i.e. a child's reactions to transitions), demandingness (for attention by intrusions and/or aggression), mood (as reflected by excessive crying, anxiety, or provoking anger), and distractibility/hyperactivity (which require high vigilance and active parental management). The other two factors include 'acceptability', which addresses how closely a child meets parental expectations (partly by possessing socially desirable characteristics), and 'parental reinforcement' which usually results of positive parent–child interactions (Abidin, 1995).

Factors constructing the parent domain include parents' personality components and parental functionality. These factors include attachment, depression, and a sense of competence in the parental role. 'Attachment' is determined by the intrinsic investment parents have in their role and their motivation to fulfill it; 'depression' as comprises parental energy and emotional availability to their child; and 'parental competence'

reflects the degree of comfort in decision making processes and disciplinary abilities. Feelings of guilt are included in this domain as well (Abidin, 1995).

The situational factors affecting parenting stress in Abidin's model are the spouse, (parental) health, isolation, and role restriction. The 'spouse' represents a role partner and a support system; parental 'health' might affect the parenting abilities; social 'isolation' or lack of social support; 'role restriction' represents the impact of parenthood on other life roles, and on a parent's sense of personal freedom. Lastly, life events occurring outside the parent–child system may moderate or exacerbate parenting stress, as they influence parental emotional resources and abilities to cope with the parenting role (Abidin, 1995).

Abidin's model guided the construction and validation of the Parenting Stress Index (PSI), which has become the parenting stress assessment-of-choice in research and clinical settings. The PSI is a standardized self-report questionnaire for parents, comprised of subscales measuring stress across the domains described above. Normal parenting stress levels would fall between 16th-80th percentiles of the PSI scores, whereas scores greater than the 85th percentile, are considered as high stress levels (Abidin, 1995).

Abidin's model and the PSI were used in the current study as the guiding conceptual framework and the assessment tool for parenting stress. A child's illness (or a child's health) was not explicitly included as one of the stressors in Abidin's model; however, it appears to play an important role in this phenomenon of interest. Research strongly correlates child's illness to parenting stress, by demonstrating high levels of parenting stress in parents of sick children (including CHD). The stress is usually captured

in multiple PSI domains (varying by the pediatric illness), emphasizing how widely an illness affects family systems (in accordance with other family stress theories; see Boss, 2001; Hill, 1958). Rearing an infant with CHD is a difficult situation; hence this study suggests that the parenting stress characteristics in parents of infants with CHD would be different than those in parents of healthy children. The parenting stress literature provides confirmatory evidence that Abidin's framework and the PSI is able to capture high parenting stress in the population of interest of our study.

Child Development

Child development refers to the biological, psychological and emotional changes occurring in human beings in the process of maturation between birth and the end of adolescence (Smith et al, 2011). Most changes occur rapidly during the early years of life, and are determined by interacting genetic and environmental factors (Tanner, 1990). For instance, during infancy growth rate is mostly determined by genes, but infants might fail reaching their full growth potential given inadequate environmental conditions (i.e. non-genetic factors such as poor nutrition, or continuous disease). The ability to assess the cognitive capacity is limited, as neonates have very few skills; however, the human learning and information-processing abilities increase until an adult-level is reached by post adolescence (Smith et al, 2011). Similarly to the physical growth, early brain development including the socio-emotional- and cognitive capacities, are dramatically influenced by environmental factors. These factors range widely and include, among the others, environmental stimuli and social interactions (Brotherson, 2005; Bowlby, 1988; Erikson, 1963).

Child development has become a source of constant interest to professionals early in the 20th century, as renowned psychotherapists and theoreticians such as Sigmund Freud and Jean Piaget introduced their perspectives about the developmental process (Baldwin, 1967). Although their theories were predominant for many years, more sophisticated frameworks were developed later (Miller, 2009). Nevertheless, they all share the mutual premises that children go through (age-fixed) developmental stages. Along these stages, children develop their motor and language skills, learn to solve problems, acquire social behaviors, and develop a sense of trust and morality (Bowlby, 1988; Erikson, 1963; Piaget, 1954; Piaget 1965a; b). More specifically, during the first two years of life, infants learn through conditioning and mimicry. At this stage, environmental stimuli and reflexive behaviors cause habituation of experiences. Parental attentiveness to infants' needs during this stage dictates the level of trust and autonomy infants gain (Bowlby, 1988; Erikson, 1963). Between the ages of 2-7 years, children acquire language skills and problem solving abilities. Until the age of 12, children develop reasoning, thinking, and understanding of abstract concepts. The last developmental stage begins at the age of 12 (adolescence) and might continue into adulthood (Erikson, 1963; Piaget, 1954; Piaget 1965a; b)

All developmental theoreticians also agree that parental practices play an important role in children's developmental processes, and especially during the early infancy (Bowlby, 1988; Erikson, 1963; Piaget, 1954; Piaget 1965a; b). Practices such as smiling to the infant, eye-contacting, stimulating, responding to the infant's needs, and showing affection importantly contribute to a healthy infant development (Bowlby, 1988;

Erikson, 1963; Piaget, 1954; Piaget 1965a; b). Attachment Theory (Bowlby, 1969; Bowlby, 1973; Bowlby, 1988), one of the most acclaimed frameworks in this field, claims that the need for parental closeness is inborn for survival and protection purposes, and it is not secondary to other physical needs. Early behaviors (i.e. crying, smiling) serve as a means for an infant to attract or signal caregivers. According to Attachment Theory and other developmental frameworks, infants who receive sensitive care become secure ('secure attachment') that their needs will be fulfilled. They can than safely explore their environment, learn, associate with others, and develop a sense of trust (Bowlby, 1988; Erikson, 1963).

Secure attachment depends on caregivers' abilities to correctly interpret infant signals and properly respond to them in a timely manner. It cannot be established without available and consistent care, or if the infant's needs are being ignored. Secure attachment serves as a foundation for healthy development, helps infants establishing high self-esteem and determines their level of trust, empathy, and aggression (Bowlby, 1988; Thompson, 2008). Children lacking secure attachment are at risk for severe emotional deficits, maladjusted behaviors, and disturbed interaction skills as they grow up (Howe, 2005; Pearce & Pezzot-Pearce, 2006; Thompson, 2008). Secure attachment serves as a framework for emotional behaviors throughout adulthood, impacting individuals' abilities to be emotionally involved in social relationships (Hazan & Shaver, 1987). Attachment theory has been criticized for over emphasizing the caregivers' role in the infant-caregiver relationship (Smith et al, 2011), while neglecting infants' temperamental aspects as contributing to the system (Thomas & Chess, 1986). However,

the most current assumptions are that parental practices and infant temperament interact and affect each other in a reciprocal manner (Vaughn & Bost, 1999).

Examination of the attachment- and the development research clearly indicates that parenting stress does interfere with both the formation and quality of parent-child relationships, necessary for fostering the optimal growth and development of infants, by altering the important parenting practices previously mentioned (Ello & Donovan 2005; Stelter, & Halberstadt, 2011; Carey, Nicholson, Fox 2002). Therefore, these developmental frameworks underlie the assumptions of the current study that increased parenting stress might be associated with infants' adverse developmental outcomes.

Congenital Heart Defects

CHD is a collective name for a group of structural abnormalities in the heart or the great vessels formatted in utero, mostly during the first gestational trimester (Ball & Bindler, 2008). It is the most prevalent congenital anomaly, occurring in approximately one percent of live births; and it is more prevalent in males than females (van der Bom et al., 2010). Defects are usually diagnosed prenatally, soon after birth, or within the first year of life (Ball & Bindler, 2008). The CHD etiology is usually attributed to the interaction of genetic and environmental factors. Such factors may include exposure to drugs during pregnancy, maternal infections or metabolic disorders, chromosomal abnormalities, and more (Ball & Bindler, 2008). The incidence of CHD in children with chromosomal syndromes, such as Down syndrome, trisomy 13 and 18, for instance, exceeds 50% (Wernovsky, 2006). CHD is the leading cause of death during infancy (excluding

prematurity), and with varying long-term survival rates (95% to 80%) by disease complexity (Marino et al., 2012). Deaths from CHD have decreased dramatically over the past several decades, even for the most complex palliated defects, due to advances in medical care, becoming one of the most common chronic illnesses of childhood (van der Bom et al., 2010). Because of the genetic component of CHD, incidence is expected to rise as the survivors have children of their own (Marino et al., 2012).

CHD encompasses more than 35 types of defects ranging in severity and prognosis (Ball & Bindler, 2008). The classification of CHD is still occasionally debated. In the past, CHD was categorized into cyanotic and acyanotic defects. Nevertheless, defects are currently classified by pathophysiology and hemodynamics as follows: defects causing (a) increased pulmonary blood flow, (b) decreased pulmonary blood flow, (c) obstructed systemic blood flow, and (d) mixing of systemic and pulmonary blood (Ball & Bindler, 2008). Defects causing increased pulmonary blood flow are the most common, resulting from a connection of blood flow between the right and left side of the heart or between the great arteries. In such defects, the higher left-heart pressure leads to left-to-right shunt, increasing the pulmonary circulation. In defects obstructing the pulmonary blood flow, the often existing right-to-left shunt causes little or no blood reaching the lungs for oxygenation resulting in cyanosis. Defects causing obstructed systemic blood flow are caused by left ventricular dysfunction or severe left outflow obstruction, decreasing cardiac output. Many complex conditions involve a combination of defects that make the infant dependent upon mixing systemic and pulmonary circulations for survival. Such defects, for example, include the Truncus Arteriosus, Transposition of the Great Arteries

(TGA), Tetrology of Fallot (TOF), and Hypoplastic Left Heart Syndrome (HLHS) (Ball & Bindler, 2008; Marino et al., 2012). Another common classification of defects determines their complexity by the surgery type (corrective vs. palliative), which eventually determines the heart's post-surgical functionality (Bi-ventricle vs. Single ventricle) (Torowicz et al., 2010).

The clinical manifestations and symptoms of CHD vary by the severity and the pathophysiology of the defect. Heart murmur, indicating high pressure blood flow through a shunt or a narrowed valve, is often the first, and sometimes the only indicator to be noticed (such as in infants with a small atrial septal defects) (Ball & Bindler, 2008). Other defects may have specific manifestations according to the nature of blood flow obstruction, and even threaten life. Additional signs and symptoms may include cyanosis shortly after birth, dyspnea, fatigue, diaphoresis, and clubbing of the fingers and toes. Along with their cardiovascular compromise, infants often display feeding difficulties (as they periodically need to stop sucking to breath) that might cause growth delays (Medoff-Cooper & Irving, 2009; Medoff-Cooper, Naim, Torowicz, Mott, 2010; Medoff-Cooper, 2011). Severe obstructions may lead to hyper-cyanotic episodes, caused by physiologic decrease in oxygen pressure. Hyper-cyanotic episodes typically occur with morning rise or suddenly following crying, warm baths or increased physical activities. Episodes are characterized by tachycardia and tachypnea, cyanosis, poor tissue perfusion to the degree of seizures and loss of consciousness. Lesions causing severe obstructions and low cardiac output are characterized by diminished pulses and delayed capillary refill time,

and might lead to decreased urinary output and even shock. Infants with the most complex CHD remain at risk for congestive heart failure (CHF) (Ball & Bindler, 2008).

Treatment for CHD depends on the severity of symptoms and whether the condition is imminently life threatening. Some conditions are self-correcting (e.g. small ventricular septal defects), whereas others require complex surgical interventions (e.g., HLHS, TGA, tetralogy of Fallot) (Ball & Bindler, 2008; Marino et al., 2012). Catheterization or surgical correction of the defect is the treatment of choice in many cases, leading to complete recovery of the child. Some complex conditions, however, allow only for palliative interventions. Surgeries are usually performed early in infancy in order to prevent the major complication of irreversible pulmonary hypertension, and to avoid secondary damage to the brain, heart, lungs, and other organs. Some surgeries may be postponed with palliative procedures until the infant has grown, potentially increasing their success. Children with complex defects might require multiple stages of palliative and corrective surgeries and revisions, valve replacements or catheterization, and even pacemaker implants. The post-operative period is followed by breathing difficulties and pain, and includes care in the intensive care unit, monitoring for infections, heart functioning problems, and other potential complications (e.g. feeding issues, poor weight gain, Post-Pericardiotomy Syndrome). Depending on the defect and treatment complexity, infants may be discharged within a few days, or may require long-term care (Ball & Bindler, 2008).

Parenting Stress in Families of Infants and Children with CHD

Parents of infants with CHD are potentially exposed to high levels of stress even before their infant is born, given the fact that CHD is often being prenatally diagnosed (as early as 20 weeks of gestation). Regardless the diagnosis timing, the initial period of hospitalization and early care of an infant with CHD at home can be very stressful for parents and other family members (Ball & Bindler, 2008). Children can be managed at home, or may need additional support and hospitalization until surgery, depending on the severity of their condition. Families are often provided with information about the condition, symptom management, treatment options, and post-operative care (Ball & Bindler, 2008). Ideally, this information should be spread over a period of time in order to allow parents to adjust and make educated decisions; however, this might not always be the case, as parents of infants with life-threatening defects must frequently make decisions quickly. Stress and anxiety are common among parents, as they fear that their infant will not survive until the surgery (Ball & Bindler, 2008). The post-operative period continues to be stressful as infants are at risk for complications months after surgery (Ball & Bindler, 2008). Parental thoughts and worries about long-term complications, and the future implications the illness might have are also present (Carey et al., 2002).

As with regard to parenting stress, studies show that parents of children with CHD experienced increased levels of parenting stress, in comparison to parents of healthy children (Goldberg et al, 1997; Visconti et al, 2002; Torowicz, et al., 2010; Uzark & Jones, 2003). In some cases, parenting stress levels in CHD groups were even higher than in other illness groups (e.g. cancer, Cystic Fibrosis) (Mullen et al, 2014; Goldberg,

Morris, Simmons, Fowler, & Levison, 1990). Review of the parenting stress research revealed numerous factors that act as sources of parenting stress for parents of children with CHD. Most of these factors align well with the parenting stress model; however, over the years, illness-related sources appear to join Abidin's factors.

Sources of parenting stress in parents of children with CHD

Illness-related stressors. The most influential illness-related factors are the severity of CHD, the diagnostic procedures, and the increased caretaking demands. Previous research demonstrated that parents of infants with more complex CHD, such as life-threatening defects and/or conditions requiring complex surgical procedures had higher stress levels than parents of infants with less complex defects (Dudek-Shriber, 2004; Torowicz et al., 2010; Sarajuuri et al, 2012; Uzark & Jones, 2003). Medical procedures and the intensive-care environment also evoked parenting stress (Dudek-Shriber, 2004; Smith, Hefley, & Anand, 2007; Carey et al., 2002). Particularly, longer hospitalizations and miscommunication with healthcare providers were dominant stressors across studies (Hayes & Knox, 1984; Smith, et al. 2007; Dudek-Shriber, 2004; Farley et al. 2007). For example, parents in Hayes and Knox's study (1984) described feelings of helplessness as a result of the constant need to seek information about their child's condition, as well as to negotiate care with healthcare providers. Another major issue with regard to the caretaking demands was infants' feeding difficulties (Carey, et al. 2002; Dudek-Shriber, 2004; Farley et al., 2007; Sarajuuri, et al. 2012; Torowicz, et al. 2010). Oral feeding is one of the expectations of a healthy infant, and when an infant with CHD does not succeed to feed by mouth this is very stressful for parents (Lobo, 1992).

Parents of infants who were fed by tube, had higher levels of parenting stress than parents of orally fed infants (Torowicz et al, 2010).

Child-related stressors. Another group of factors contributing to the parenting stress in this population was related to the children. Children's temperamental characteristics (Carey, et al 2002; Torowicz, et al. 2010; Uzark & Jones, 2003), and behaviors (Darke & Goldberg, 1994; Dudek-Shriber, 2004; Visconti et al., 2002; Young Seideman et al., 1997) were often strongly related to parenting stress. For instance, Torowicz et al (2010) found that the demandingness of children varied according to their cardiac pathophysiology, resulting in higher stress levels in parents of more demanding children. Similarly, parents who perceived their children as irritable, having negative mood, and/or being difficult to sooth, had higher parenting stress levels than parents who did not perceive their children's temperaments as such (Carey et al, 2002; Torowicz et al., 2010). Across studies, parents expressed a desire for normalization of their children (Carey, et al. 2002; Hayes & Knox, 1984). Levels of stress were influenced by parental perceptions of the appearance of their child (Dudek-Shriber, 2004; Young Seideman et al., 1997), parental acceptance of the illness (Carey, et al. 2002; Darke & Goldberg, 1994; Uzark & Jones, 2003), and by the reinforcement parents receive from their children (Darke & Goldberg, 1994). Studies reported inconsistent results regarding associations between child's age and parenting stress (Farley et al., 2007; Uzark & Jones, 2003).

Parent-related stressors. Various parental characteristics such as low education level, unemployment status, younger age and female gender also predicted stress (Darke & Goldberg, 1994; Dudek-Shriber, 2004; Goldberg, et al., 1990). Parents who

experienced high levels of parenting stress also frequently experienced emotional distress, uncertainty, and depression (Carey, et al. 2002; Darke & Goldberg, 1994; Farley et al., 2007; Goldberg, et al., 1990; Lawoko & Soares, 2002). Issues of attachment, and communication problems (with the child) commonly predicted high parenting stress levels (Darke & Goldberg, 1994; Dudek-Shriber, 2004; Goldberg, et al., 1990; Hayes & Knox, 1984; Young Seideman et al., 1997). A low sense of competence affected parental abilities to set limits or discipline their child (Darke & Goldberg, 1994; Goldberg, et al., 1990; Young Seideman et al., 1997; Carey et al, 2002; Uzark & Jones, 2003).

Family and social-related factors. Disruption of the normal family life was a major concern with regard to increased parenting stress. Parents reported difficulties in addressing other familial needs, were worried about, and felt guilt towards the other family members (Carey, et al., 2002; Farley et al., 2007; Hayes & Knox, 1984). Parents who were separated, divorced, or had poor- quality marriage, scored higher on parenting stress measures (Dudek-Shriber, 2004; Rimmerman & Stanger, 2001). Lack of financial resources, little social support, and poor ability to manage the illness were predictive of high stress levels (Dudek-Shriber, 2004). Some studies demonstrated beneficial effects of social support on parenting stress relief (Phipps & Drotar, 1990; Visconti, et al., 2002). Several other studies, however, found no significant relationships between socio-familial characteristics (e.g. marital or socioeconomic status, family coping) and parenting stress (Farley et al, 2007; Uzark & Jones, 2003).

Development of Infants and Children with CHD

Most infants and children with CHD [without co-existing Central Nervous System (CNS) abnormalities at birth] develop normally. As a group, however, they have a significantly higher incidence of neurodevelopmental delays, compared to the general population (Ballweg et al., 2007; Wernovsky, 2006; Marino et al., 2012). The incidence and severity of the impairments (i.e. mild or combined disabilities, severe impairment) vary according to the defect type and complexity (Marino et al., 2012). These neurodevelopmental delays include cognitive and intellectual impairments, and are characterized by tone and motor abnormalities, feeding difficulties, problems with visual-motor integration, and speech delays through infancy. Later in childhood, developmental delays can appear in the form of executive function deficits, learning difficulties, behavioral abnormalities, inattention and hyperactivity. These delays can last as children progress through school, leading to long-term implications such as academic failure, poor social skills, low self-esteem, and behavioral disinhibition (Ballweg et al., 2007; Wernovsky, 2006; Marino et al., 2012). The causes of these delays are multifactorial and include factors specific to the child, the illness, and the environment.

Child-Related Factors

Some genetic syndromes have CHD and neurodevelopmental delays as part of their clinical phenotype. Such syndromes include trisomies 13, 18 and 21; DiGeorge, Williams, and Noonan's syndromes; APOE genotype; and CHARGE association (Ballweg et al., 2007; Gaynor et al., 2003; Wernovsky, 2006). These genotypes or chromosomal abnormalities have a high incidence of CHD among multiple other

congenital abnormalities, and are independently and nearly always associated with (in many cases, severe) neurodevelopmental delays (Ballweg et al., 2007; Wernovsky, 2006).

Similarly**,** increasing evidence shows that in-utero CNS-development is abnormal in fetuses with CHD. The nervous and cardiovascular systems form nearly simultaneously in early gestation; therefore, abnormalities in one system increase the likelihood of having problems in the other (Wernovsky, 2006). Factors affecting the CNS-development mostly relate to low cerebral blood flow (CBF), and impaired brain CO_2 reactivity. These factors are often evidenced by white matter injury (Periventricular Leukomalacia, PVL), and microcephaly (Brown, Wernovsky, Mussatto, Berger, 2005; Ballweg et al, 2007; Wernovsky, 2006). Furthermore, these factors are associated with neurodevelopmental delays and a higher risk of death in all age groups (Ballweg et al, 2007). Additional birth risk factors that predict neurodevelopmental delays in children with CHD (and in the general pediatric population) are prematurity, lower birth weight and birth head circumference, and lower Apgar Scores (Ballweg et al, 2007; Galli et al, 2004; Trittenwein et al, 2003; Jonas et al., 2003; Gaynor et al., 2006). The association between prematurity and cognitive development varies with the degree of prematurity, so that younger gestational age is associated with worse developmental outcomes (Voigt et al., 2013).

Though infants with CHD are usually born with normal weight for gestational age (Nydegger & Bines, 2006), many experience growth problems (stunting) early in life, which might later affect their neurodevelopment (Chang, Walker, Grantham-McGregor,

Powell, 2010). Malnutrition is the leading cause of growth problems among infants with CHD, and is usually attributed to the feeding difficulties they experience, and their increased metabolic rate. The complexity of CHD is directly related to likelihood of nutritional implications (Nydegger & Bines, 2006; Steltzer, Rudd, Pick, 2005). Many infants with a small defect would not have any feeding problems. However, infants with more complex defects might be unable to take in enough calories, due to sucking coordination difficulties, and increased heart and respiratory rates during feedings. Weight gain rate is affected more than height gain rate, but complex defects (Single-ventricle physiology) causing severe illness or continuous malnutrition can cause linear growth stunting (Ravishankar et al., 2013). Among the other factors causing growth delays are malabsorption, prolonged hospital stays, and postoperative complications (e.g. fever, sepsis). Up to 50% of the infants with complex CHD are diagnosed with growth failure or failure to thrive (FTT) (Peterson & Wetzel, 2004) at some point during infancy. FTT describes a clinical syndrome of decreased growth (less than the fifth percentile on a standardized infant growth chart) or static/slow growth rate over a period of months (Ward, Lee, Lipper, 2000). Malnutrition and FTT in infants with CHD relate to both physical and cognitive delays, affecting brain development, oral-motor skills, and social skills (Chang et al., 2010; Nydegger & Bines, 2006; Steltzer, Rudd, Pick, 2005).

Illness-Related Factors

CHD diagnostics, operational, and post-operational factors, were all identified as predictors of neurodevelopmental delays, mostly attributed to neurological injury caused by abnormal fetal cerebral physiology, hypoxemia, hypotension, and low cardiac output

(Brown et al., 2005). The CHD diagnosis is a strong predictor which corresponds to the incidence and the severity of the developmental deficits. Only the minority of children with milder defects (e.g., ventricular septal defect) have mild neurodevelopmental abnormalities. The incidence, however, increases with the complexity of the defects (e.g., TGA, anomalous pulmonary venous) to the degree of which only the minority of children with extremely complex defects (e.g. HLHS, non-HLHS functional univentricular heart) are normal in all developmental respects (Wernovsky, 2006).

The diagnostic severity determines the course and management of the illness, which eventually affect the degree of the neurological injury. For instance, prenatal diagnosis of complex defect allows for early initiation of prostaglandins to maintain the ductus arteriosus open, preventing the acidosis causing neurologic injury (Ballweg et al, 2007). Studies showed associations between the timing and type of cardiac surgery and later neurodevelopmental outcomes. Such surgeries for example, involved cardiopulmonary bypass (CPB) technologies with deep hypothermic circulatory arrests (e.g., Fontan operation) (Wernovsky, 2006; Ballweg, et al., 2007; Goldberg et al., 2000; Newburger et al., 1993). More recent studies, however, demonstrate no such associations, as more advanced technologies and surgical techniques are being applied (Fuller et al., 2009; Marino et al., 2012). Several post-operative factors such as seizures, early reoperations, mechanical ventilation support, and prolonged/multiple hospitalizations are considered as risk factors as well (Bellinger et al., 1995; Fuller et al., 2009; Wernovsky, 2006). The effect of length of hospital stay on later neurodevelopmental delays appears to remain strong even after controlling for other post-operative events (Fuller et al., 2009;

Newburger et al., 2003). The dominance of the length of hospital stay over other clinical predictors might be explained by the fact that it is determined by many illness-indicators such as hypoxemia, hypotension, arrhythmia, sepsis, and PVL (Ballweg et al., 2007).

Social and Familial (Environmental) Factors

Socioeconomic status (SES) is perhaps the strongest predictor of neurodevelopmental outcomes (Wernovsky, 2006). Parental age, intelligence, education, occupation, and income are closely correlated with developmental outcomes in children with CHD (Ballweg et al, 2007; Davis-Kean, 2005; Fuller et al., 2009; Wernovsky, 2006). Other social determinants of health, such as gender, race, and ethnicity were studied to a lesser degree (Ballweg et al, 2007). Nevertheless, several studies identified differences between male and female infants, indicating male infants to be less communicative and less responsive to stimuli than female infants (Weinberg, Tronick, Cohn, & Olson, 1999).

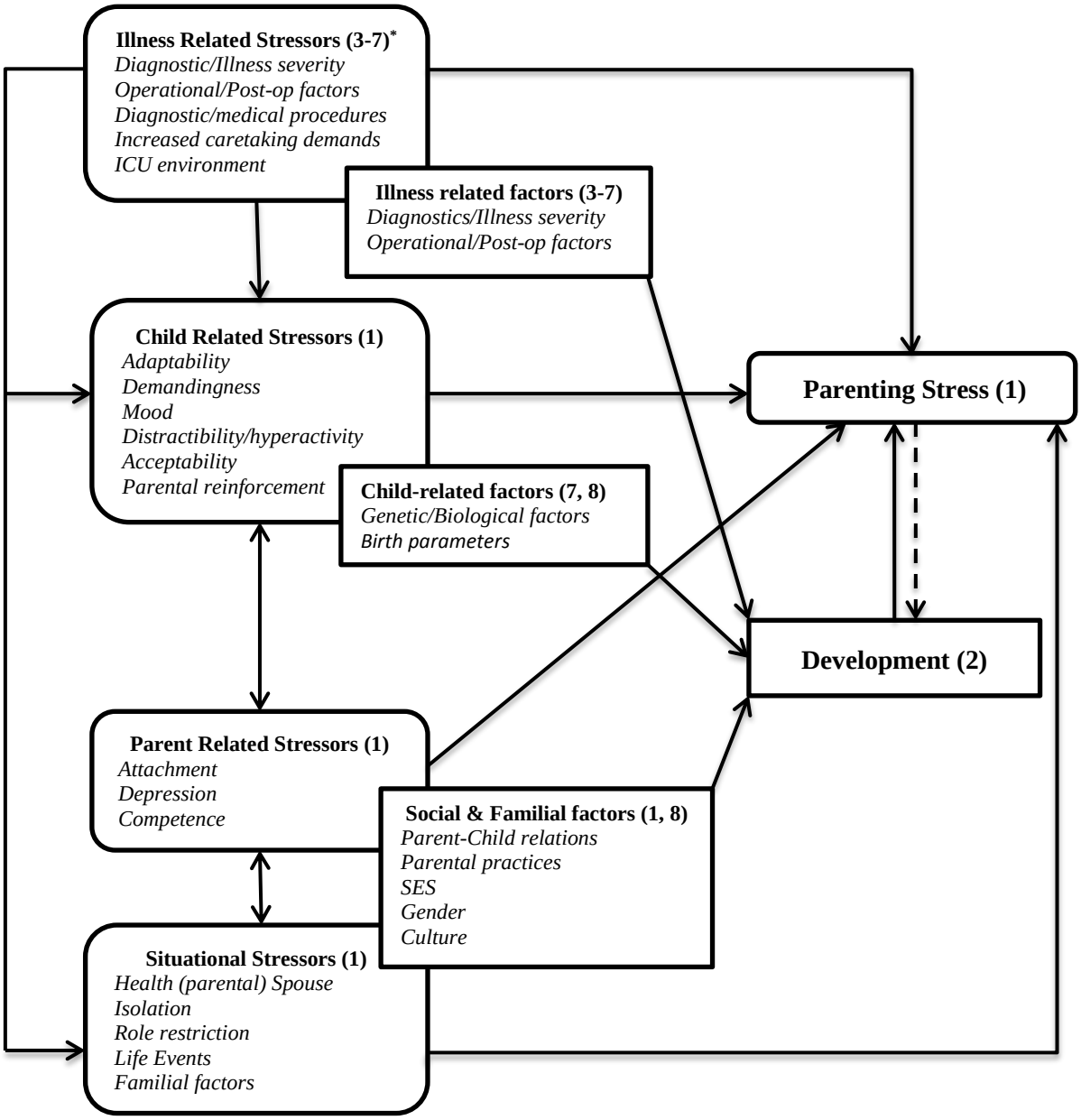

Figure 2: A conceptual Model for the Proposed Study

**Numbers are linking the model components to the study variables, as are presented in Table 1 [e.g. (1) = PSI subscales in Table 1; (8) = Demographics in Table 1, etc.].*

The conceptual model for the study summarizes the components constructing the concepts of interest, as identified by the previously mentioned theoretical

frameworks, and by previous research. Most of the stressors were adopted from Abidin's original model; however, additional illness-related stressors and familial stressors were mentioned in the literature, and therefore, were also included in the model. Similarly, the factors affecting development were gathered from the literature and from traditional theories of development. The explored associations between parenting stress and infant development are represented by the broken arrow.

Gaps

Review of the CHD literature demonstrates that parents of children with CHD experience increased levels of parenting stress on standardized parenting stress measures. Nevertheless, no evidence exists with regard to the parenting stress change over the first year of infants' life. Studies concluded that longitudinal research is needed, in order to better understand what happens to parenting stress over time (Uzark & Jones, 2003). The current study longitudinally investigates the change in parenting stress over the critical period of infancy, during which most medical interventions occur, and during which parents adjust to, and learn how to cope with their infant's illness.

Furthermore, parenting stress is associated with adverse neurodevelopmental outcomes and behavior problems among diverse pediatric populations. Positive associations between parenting stress and neurodevelopmental delays were found in healthy infants (Molfese, et al., 2010), preterm infants (Grunau et al., 2009; Voigt, Brandl, Pietz, Pauen, Kliegel, & Reuner, 2013), and children with disabilities other than cardiac disabilities (Hintermair, 2006; Neece, Green, Baker, 2012). Surprisingly, only a handful of studies examined similar associations in children with CHD (Goldberg, et al. 1991;

DeMaso, Beardslee, Silbert, Fyler, 1990; DeMaso, et al., 1991), despite their increased incidence of developmental delays (Wernovsky, 2006; Marino, 2012). These relationships were demonstrated mostly by cross-sectional analyses in children at various ages, and posited a transactional effect on each other. Such analyses are limited in their ability to provide temporal perspective for the effect of stress on development.

Nevertheless, longitudinal studies in other pediatric populations provided temporal evidence for the effect parenting stress has on children's neurodevelopment. For instance, Goldberg et al (1997) found that parenting stress over the first three years of life is the strongest predictor of child behavior problems at the age of four years. Similarly, Grunau, et al, (2009) showed that higher parenting stress exacerbated the effect neonatal pain had on cognitive outcomes of preterm infants at 18 months of age. Such temporal associations, however, have not yet been demonstrated in infants with CHD. The current study can potentially provide temporal explanations, by analyzing associations between earlier parenting stress and later neurodevelopmental outcomes.

CHAPTER 3: METHODS

Introduction

Chapter three discusses the design and methodology of the proposed study. As this is a secondary analysis to a prospective cohort study, the original objectives, settings, and procedures of the parent study are described in detail. The methodology for the study is presented, followed by a description of the variables and their measures. Next, the analysis plan is discussed according to the study's aims, including the handling of missing data. Finally, the study's ethical and human-subjects considerations are discussed.

Methods

Study Design

The current study was a secondary analysis of data obtained from a larger prospective cohort study. The parent study named *Feeding Behaviors and Energy Cost in Infants with Congenital Heart Disease* (NIH/NINR R01 NR002093; MO1-RR00240; UL1-RR-024134), was conducted during 2003-2007. The study originally examined feeding aspects predicting FTT in infants with CHD, such as feeding performance, energy expenditure, and other growth parameters. Infants were examined during five visits over their first year of life: at their first six weeks of life as newborns; at three months; at six months; at nine months; and at twelve months of age.

Setting and Participants

The parent study was conducted at The Children's Hospital of Philadelphia (CHOP), a 480-bed general facility serving Pennsylvania and New Jersey areas (US

News & World report, 2015). The parent study included a convenience sample of infants with CHD, and a control group of healthy infants. The CHD sample was recruited from the cardiac intensive care unit (CICU) of the Cardiac Center at CHOP, one of the largest cardiac centers in the country, performing over 1,200 catheterizations and 600 open-heart surgeries yearly (Cardiac Center CHOP, 2014). The healthy sample was recruited from CHOP primary care practice, the faculty practice, and the community. Inclusion criteria for the CHD group were as follows: infants with complex CHD, who underwent corrective or palliative surgery for their heart defect within their first six weeks of life; and who were without other congenital anomalies other than their cardiac defect or other documented genetic syndromes (except 22q deletion and DiGeorge syndrome). Infants born <35 gestational weeks, and/or weighed < 2000 grams at birth were excluded, as were infants with other congenital or acquired lesions, potentially effecting feeding, growth, or development (e.g. gastrointestinal disorders, orofacial clefts, neurological impairments). A total of 33% of the CICU infants were enrolled during the study period. The enrollment rate resulted from parents refusing to consent, parents unwilling or unable to return for follow-up, and other studies simultaneously conducted in the CICU competed for enrollment. Demographic characteristics of unenrolled infants (with CHD or healthy) are unavailable. The total enrollment for both cardiac and healthy groups was 241 infants. The entire sample of both the CHD and the healthy group was used in the current study. The final sample size for each study aim was determined by the attrition rate, and by the missing reporting of values within the variables of interest.

Study Procedures and Data Collection

The study was approved by the Institutional Review Board of CHOP, Philadelphia, PA. Infants were post- surgically screened for eligibility, and parents of eligible infants were approached for enrollment. Informed consent was signed by parents or legal guardians of all enrolled participants. Data were obtained at discharge, and during subjects' outpatient visits at CHOP: Assessments of infants' vital signs were obtained at the General Clinical Research Center (GCRC) by nurses; growth and other study variables were assessed by trained research staff, at the Nutrition and Growth Laboratory (NGL). Parents were requested to fill in self-reporting questionnaires including demographic information, infant temperament, and parenting stress; and to record infant's diet close to their visits. Parents received meal and parking vouchers for their infant assessment visits, and gift cards upon the return of the questionnaires.

Visits varied by assessment time (ranged between 30min-5 hours), and by the variables measured. The newborn visit included measures of feeding performances (e.g. suck-swallow-breathe coordination), anthropometrics (i.e. weight, length, head circumference, and skin folds measurements), and body composition. These measures were included in the three-month visit as well, in addition to measures of infant temperament, parenting stress, and energy expenditure. The six-month visit included feeding and anthropometrics measures, parenting stress, infant neurocognitive development, and temperament. The shortest nine-month visit included only anthropometrics and parenting stress measures. The last twelve-month visit included anthropometrics, energy expenditure, parenting stress, infant neurocognitive development, and temperament. Additionally, infants' diet and medications were

recorded at each visit. The current study used only certain measures from the parent study, which are necessary to examine the change in parenting stress over time, and to determine relationships between parenting stress and infant development.

Study Variables and Instruments

Parenting stress. Table 1 presents the study variables and the measures used to assess them. Parenting stress was assessed at three-, six-, nine-, and twelve-month visits, via the Parenting Stress Index (PSI)-Long Form (Abidin, 1995). The PSI is a standardized, self-reporting questionnaire designated for parents, measuring stressors in the domains identified in Abidin's model (Child Domain, Parent Domain, and Life Stressors). The PSI-Long Form consists of 120 items, yielding scores over 17 subscales. Forty-seven items ranked on a 5-point Likert style scale (1=strongly agree, 5= strongly disagree) measure Adaptability, Acceptability, Demandingness, Mood, Distractibility/Hyperactivity, and Reinforces Parent. These six subscales construct the Child Domain. An example item is "My child seems to cry or fuss more often than most children". Fifty-four, 5-point Likert style scale items, measure Depression, Attachment, Role Restriction, Sense of Competence, Isolation, Spouse, and Parent Health. These seven subscales construct the Parent Domain. Item for example: "I often feel guilty about the way I feel toward my child." Independent scores from the Parent and Child Domains are summed to an overall score, and construct the Total Stress subscale. An optional subscale measuring the Life Stress Domain lists 19 stressful life events, potentially experienced by parents outside the parent-child system (e.g. divorce, income decrease,

troubles at work, etc.). Parents' yes/no responses are summed to an index, so that a higher total Life Stress score indicates more situational stress (Abidin, 1995).

Interpretation of the PSI scores can be mainly based on the Total Stress, higher scores thus indicating higher parenting stress levels. The normative PSI scores fall between the16th-80th percentiles; low stress levels fall beneath the 16th percentile; and any score at, or above the 85th percentile is considered high (Abidin, 1995). Abidin suggests that a Total Stress score over 260 indicates that parents might be at risk for adverse outcomes. He further suggests that high Life Stress scores tend to intensify the Total Stress; therefore, a Life Stress score over 17 buffers the Total Stress score. Individual subscales' scores can be also interpreted independently, thus allowing to analyze specific aspects of the parent-child system.

The PSI was validated on demographically diverse samples of 2,663 mothers and 200 fathers, who had children from 1month to 12 years of age. Alpha reliability coefficients for the different subscales of the Parent Domain range between .70 -.84; for the Child Domain range between and .70-.83; and for the Total Stress score is above .90. Test-retest reliability between one- to three-months interval for the Child Domain, Parent Domain, and Total Stress were .63, .91 and .96, respectively (Abidin, 1995).

Infant development. Infants' neurocognitive development was assessed at six and twelve months of age, by doctoral level developmental psychologists, using the Bayley Scales of Infant Development-2nd Edition (BSID-II; Bayley, 1993). During the parent study period, the BSID-II was the most widely applied, standardized measure (the revised BSID-III was only introduced in 2005), used to assess children's neurocognitive

development between 1-42 months of age (Nellis & Gridley, 1994). The BSID-II consists of three scales: The Mental Scale, constructed of 178 items measuring mental capacity and cognitive skills, such as coordination, language acquisition, and problem solving skills; the Motor Scale, constructed of 111 items measuring gross and fine motor skills; and the Behavior Rating Scale (BRS), constructed of 30 subjective-rating items forming four criteria to assess the child's emotional, behavioral, social, and environmental orientation. The number of items used for scoring change according to the examinees age. For example, at the age of 1-2 months, infants are tested for only 9 Motor Scale items (Bayley, 1993). Child's testing position (e.g. seated, supine), materials needed for testing (e.g. shaped cubes fitted into a form board), and related items are instructed by the test protocol.

The raw Mental and Motor scores are transformed into the Mental Development Index (MDI), and the Psychomotor Development Index (PDI), respectively. The MDI and the PDI were standardized on a normative sample of 1,700 infants, with scores ranging between 50 and 150 (mean=100, SD=0.15; Bayley, 1993). Additionally, the Mental and Motor scales' items can be sorted into a Motor, Cognitive, Language, and Personal/Social "facets," so that the child's developmental age could roughly be estimated (no clear cut-off point). The BRS produces (subjective) scores for the age relevant criteria, and a total percentile ranking score (Bayley, 1993).

Alpha reliability coefficients of each scale during the measure development were calculated within various age groups, and ranged between .64- .93. The lowest coefficients were among the youngest groups on the BRS, and among the highest age

levels on the Motor scale. Test-retest reliabilities measured over a period of 16 days were relatively high for the Mental and the Motor Scales (r=.83-.91 and .77-.79; respectively), and intermediate to high (.48-.90) for the BRS (Bayley, 1993). A retest reliability of the BRS also determined 73.3-96.5 percentage of agreement across two tests, with lowest reliability at the lowest age level (1 month). Inter-rater reliability coefficients for the Mental and Motor Scales were .96 and .75, respectively. For the BRS subscales, these coefficients ranged between .57 -.83 (Bayley, 1993).

Content validity of the three scales was established by a panel of over 25 experts. For the Mental and Motor scales it was determined by deriving correlation coefficients of the items and the scales (i.e. mental vs. motor) and facets (i.e. cognitive, language, social) in which they were placed. The correlations between the MDI and the PDI were reported to be moderate to low (Bayley, 1993). Most items correlated with the facet in which they were placed; or replaced into the facets with which they were highly correlated. The Social facet seems to be poorly covered, with only 19 items, and only three items beyond the age of nine-months (Bell & Allen, 2000). Construct validity for the BRS was established by factor analyses, and by examining correlations between the BRS and the other two scales. Predictive validity is not addressed by the test developers. Concurrent validity indicated moderate to high correlations when compared to various ability and language tests, such as the Differential Abilities Scale (DAS; Elliot, 1990), and the Preschool Language Scale-3 (PLS-3; Zimmerman, Steiner & Pond, 1992). Finally, the clinical validity was determined in a series of studies examining eight groups of clinical

pediatric populations (e.g. premature, Autistic). Almost all groups performed significantly below the mean (Bayley, 1993).

Covariates considered in the analyses

Severity of illness. The literature review demonstrated that severity of illness is related to both parenting stress and neurodevelopment of children in the general pediatric population, and in populations of children with CHD. While several assessments were used to represent the degree and severity of infants' illness in the parent study, the following measures were considered to be included in the current study:

Risk for in-hospital mortality. The surgical complexity and the primary risk for post-surgical mortality were assessed in the parent study via the Risk Adjustment for Congenital Heart Surgery 1 (RACHS-1; Jenkins et al., 2002), and the Aristotle Basic Complexity Score (Lacour-Gayet et al., 2004). Since the RACHS-1 scores were reported to be stronger predictors of these outcomes than the Aristotle Score (Al-Radi et al., 2007; Kang, Tsang, Elliott, de Leval, Cole, 2006), only the RACHS-1 was considered in the current study. The RACHS-1 was developed by a panel of experts, in a consensus-based process, and has six-risk categories representing the complexity and the mortality risks from 1-6 on an ordinal scale (6 being the highest risk). Each category represents a group of procedures that carries similar risks for in hospital mortality. Defects requiring multiple surgical procedures are usually categorized into the riskier categories (as are other complex and palliative procedures). The mortality rates by category, computed from two datasets which were used for the measure's validation process, were as follow: 0.4% in risk category 1; 3.8% in category 2; 8.5% in category 3; 19.4% in category 4;

and 47.7% in category 6. An inadequate sample size in Category 5 prohibited from obtaining valid mortality rates (Jenkins et al., 2002). Since risk categories are stratified by cardiac diagnosis, the current study did not account for the infants' primary cardiac diagnosis in the analysis. The distribution of defects in the parent study's sample is presented in Table 2. The risk categories were dichotomized into low (≤3) and high (4≥) scores, as others have indicated (Costello et al., 2010).

Postoperative cardiac physiology. The RACHS-1 developers recommended adjusting for baseline risk differences in groups of children with CHD, in order to allow for meaningful comparisons of illness severity (Jekins et al., 2012). Hence, additional classification of the infants according to their post-operative cardiac physiology was performed in the parent study. Infants were categorized either into a Single-ventricle (SV), or into a Bi-ventricular (BV) physiology group (see Table 2), according to their cardiac functionality assessed by a pediatric cardiologist. Assessments were based on infants' pre- and postoperative echocardiograms, in accordance with the established standards (Friedman, Kleinman, Copel, 2002; Khairy, Poirier, Mercier, 2007).

Length of stay. Length of hospital stay (LOS) is widely associated with developmental delays in pediatric populations across studies. As LOS is often determined by other post-operative factors such as infections, PVL, seizures, hypotension, reoperations, hypoxemia, arrhythmia (Ballweg et al., 2007; Wernovsky, 2006), it is perceived as a general illness indicator serving as a proxy for these factors. Information regarding LOS in days was acquired from the infants' hospital charts.

Feeding mode. Infants with CHD may experience feeding difficulties caused by compromised sucking/swallowing/breathing coordination. These feeding issues are often associated with other illness parameters and energy imbalance, and might potentially disturb proper growth and development (Jackson & Poskttt, 1991; Medoff-Cooper & Irving, 2009). Therefore, enteral feeding supplements are commonly provided to these infants through nasogastric or gastric/jejunal tubes until they develop their oral feeding skills (Medoff-Cooper & Irving, 2009; Steltzer, Rudd, Pick, 2005). In the parent study, enteral feeding modes were recorded at hospital discharge, and at the three-month visit. Feeding modes were classified to exclusively oral feeding mode (breast or bottle), or to device-assisted feeding mode (tube only/oral and tube combined). Feeding devices included gastric, nasogastric, and jejunal tubes.

Infant growth. Infants' growth directly reflects their nutritional status. Infants who undergo surgical repair for CHD often experience sub-optimal growth (Medoff-Cooper et al., 2010; Medoff-Cooper, 2011). Poor growth might cause parents excessive stress, and can affect infant development. The proposed study will use the weight, length, and head circumference anthropometric measures collected in the parent study to assess infants' growth, as recommended by the World Health Organization (Vincenti, 1996). Measurements were obtained at all visits, by the Nutrition and Growth laboratory technicians as follows: The average of three weights measured on an infant digital scale (Scaletronix, White Plains, NY) accurate to 1 gram; the average of three length measurements obtained on an infant length board (Holtain, Crymych, UK) accurate to 0.1 cm; and the average of three head circumference measurements obtained with a

measuring tape (McCoy, Maryland Heights, MO) accurate to .1 cm. Further, measurements were converted to weight, length, and head circumference standardized z-scores, according to the WHO child growth standard charts (WHO, 2006; 2007).

Demographics. Demographic characteristics of the infants and the parents were collected in the parent study from the medical records and via parents' self-reporting. The current study considered race and ethnicity, infant gender, birth weight and head circumference, gestational age, and the presence of prenatal CHD diagnosis, as potential confounders. Additional socio-economic data (i.e. parental age, education, and income) were obtained retroactively from only a small part of the sample in the parent study; therefore, this information could not be considered for the current study. Although the parent study was not specifically directed to mothers, these were the primary participants (the sample included only two fathers). This situation derived from the fact that within the enrolled sample, mothers were those who mostly stayed with the infants during the hospitalizations, and brought them to the follow-ups.

Data Analyses

All statistical analyses were executed in STATA 13 statistical software package (StataCorp. 2013). Data from all visits were merged into a single, inclusive dataset. Missing observations and extreme values were inspected and verified through the original records. Distributions were examined to determine a need for normality transformations or variance stabilizations. Outliers were visually inspected and assessed for accuracy. Bivariate correlation matrices were generated to estimate correlations among the study variables.

Data analysis for Aim 1. In order to describe levels of parenting stress, distributions of all 17 PSI subscales at each time point were separately presented for the subjects and controls groups. Descriptive statistics included measures of central tendency and variation (mean, median, standard deviation, minimum and maximum values, and ranges) by group. Sub-group comparisons of parenting stress were performed (at each time point) using two-sample t-tests, in order to identify differences in parenting stress means at the 0.05 significance level. The underlying t-test assumptions are that groups have normal distribution and homogeneous variances (Fay & Proschan, 2010); therefore, PSI distributions in each group were tested for normality and for homogeneity (Levene, 1960; Shapiro & Wilk, 1965).

Data analysis for Aim 2. A linear mixed-effects model analysis was performed in order to examine changes in parenting stress over time (Laird & Ware, 1982; Longford, 1987). Mixed-effects models account for the fixed effects (i.e subject related factors) and the random affects (i.e. time); hence estimating the within- and between-subject variation in the outcome of interest. The estimates produced are also robust to missing data and dropout (under MAR and MCAR assumptions; Hedeker & Gibbons, 1997). All 17 parenting stress outcomes (PSI subscales) were separately regressed over the independent variable of time (represented by infants' visit from 3-12 months). The covariates that were included in the analysis were identified (from the list of potential covariates; see Table 1) in bivariate main effects models, and in bivariate two-way *covariate x time* interaction terms models. For each outcome, an initial multivariable mixed-effects model was constructed from the interaction terms which were significant at

the 0.2 level in the bivariate models. Then, the least significant interaction terms were sequentially eliminated, until only interactions significant at the level of 0.2 remained. Next, the remaining covariates, that were significant in the bivariate main effects models, were added to the multivariate models. A backwards deletion process to the final model was performed again, remaining with only significant covariates at the 0.2 level. Covariates were checked for multicollinearity ($r \geq 0.6$). Comparison of the parenting stress changes over time between the two groups relied on the *physiology x time* interaction term estimates. Differences were graphically displayed.

Data analysis for Aim 3. Associations between parenting stress (PSI subscales), and infant neurodevelopment (MDI and PDI) were examined using general linear regression models (Cohen & Cohen, 1983). MDI and PDI at six months were separately regressed over PSI subscales at three and six months; outcomes at twelve months were separately regressed over PSI subscales at three, six, nine, and twelve months. The covariates included in the analysis were identified (from the list of potential covariates; see Table 1) in bivariate models. Each outcome was separately regressed over covariates; those who are significant at the 0.2 level were included in the final multivariate models. Covariates were checked for multicollinearity. A sub-group analysis for the subjects and controls was performed.

Missing Data

Missing data patterns are important to examine in order to avoid bias and determine the most appropriate analysis plan. Therefore, patterns of missing data were examined; and estimates from the complete data analysis were compared to the estimated

generated from an imputed dataset. No consistent differences were observed between the estimates, therefore it was safe to assume no systematic bias in a form of missing not at random (MNAR; i.e. nonresponse is dependent on covariate values; Rubin, 1976).

Ethical Conduct of Research and Human Subject Considerations

The parent study and the current study were approved by the Institutional Review Board of CHOP and the University of Pennsylvania, and the research team members received the periodic, required training regarding the responsible conduct of research from the University of Pennsylvania. Parents were consented and informed regarding the study aims and procedures, potential risks and benefits, and their rights to withdraw participation without any consequences. Parents were financially compensated for their time investment. The current study involved only a secondary analysis of existing de-identified data, and therefore, possessed no risks or burden for the populations involved. No immediate benefits are expected for the study population; rather, potential benefits may apply for future pediatric populations in that findings may contribute to the body of knowledge regarding child development and parenting stress over time during the sensitive period of infancy.

In the current study, participants' privacy was maintained by using only the identification numbers assigned during the data collection process. All identifiable information was removed from the electronic data files during the parent study prior to any data analyses. The original identifiable information was stored in secured servers of the University of Pennsylvania and CHOP, accessible only to the research team members. In order to maintain data confidentiality, the identifiable data was not accessed nor used.

The current study involves no human subjects, since it was a secondary analysis. No risks were anticipated to the vulnerable populations of women and children in the parent study due to its observational nature. Although the parent study was directed towards both parents, the majority of subjects were mothers because they visited the hospitalized infants most frequently and brought them for follow-up visits. Two fathers participated in the study. As for the infants, the sample included both males and females, at an expected gender distribution similar to the CHD infant population (Marelli, Mackie, Ionescu-Ittu, Rahme, Pilote, 2007). Efforts were made to enroll eligible infants from diverse racial, ethnical, and socio-economic backgrounds.

CHAPTER 4- RESULTS

Characteristics of the Study Sample

Descriptive statistics of the demographic and clinical characteristics of the study sample are displayed in Table 3. The final sample included 167 infants, mostly white (n=134; 80%), non-Hispanic (n=119; 71%), males (n=109; 65%). The sample included 97 (58%) infants with complex CHD, of whom 49 (51%) had SV post-op physiology, and 50 (52%) had high RACHS-1 score of $\geq$4 (For detailed distribution of the defects see Table 2). The mean gestational age for all infants was 39±1.5 week, with a mean birthweight of 3365±485 grams (Table 3). The sick infant group had a median LOS of 15 (2-159) days. Among the sick group, forty (41%) of the infants were exclusively orally fed at hospital discharge, whereas 36 (37%) required device-assisted feeding (i.e. nasogastric tube, gastric tube) combined with oral feeding. At three months of age, 10 (10%) infants continued to require device-assisted feeding. Weight-, length-, and head circumference-for-age Z-scores at 3 months were -.76±1.22, -.42±1.36, and -.16±1.27, respectively. At six months of age, the mean MDI and PDI scores were 95 ±10 and 87±15, respectively. Similarly, the mean MDI and PDI scores at 12 months were 95±11 and 85±17, respectively. About half of the mothers in the sample (54%) had reported they had college degrees or greater. Among mothers of infants with CHD, fifty (52%) had received a prenatal diagnosis of their infant's CHD.

Results According to the Study Aims

Aim 1. The objective of Aim 1 was to describe and compare levels of parenting stress between parents of infants with CHD and parents of healthy controls at three, six,

nine, and twelve months of infant's life. Table 4 displays the descriptive statistics for the PSI subscales, and the PSI means' t-test comparisons between the subjects and controls groups. At three months of age, parents of infants with CHD had significantly higher PSI mean scores than parents of healthy controls on the Demandingness (18.61 vs. 14.95; $p<0.001$), Mood (9.58 vs 8.43; $p= 0.024$), Child Domain (95.52 vs. 88.51; $p= 0.040$), and Competence (24.17 vs. 22.10; $p= 0.039$) subscales. The higher Demandingness subscale mean scores, experienced by parents of infants with CHD compared to parents of healthy infants, remained highly significant throughout the entire follow-up period- at six (17.52 vs. 15.07; $p= 0.006$), nine (18.16 vs. 15.26; $p= 0.001$), and twelve (17.86 vs. 15.38; $p= 0.002$) months of age. Additional significant differences were found in the Life Stress subscale mean scores at twelve months of age. Parents of infants with CHD had significantly higher mean scores than parents of healthy infants on this subscale (10.13 vs. 7.2; $p= 0.033$).

Aim 2. Aim 2 sought to identify changes in stress levels in parents of infants with CHD over the first year of infants' life, and compare them to those of healthy controls. Tables 5a-5d summarize results from Mixed Effects regression models for PSI subscales scores over time. Each table represents a set of models, in which each PSI subscale is regressed over the independent variable of time (represented by infant follow-up visit at three, six, nine, and twelve months of age). Models in Tables 5a and 5b are adjusted for infant length Z-scores and birthweight. Models in Tables 5c and 5d represent a sub-analysis of the subjects group (by post-op cardiac physiology), and are adjusted for infant length, birthweight, and feeding mode at discharge (exclusively oral feeding vs. device

assisted feeding). All covariates included in the multivariable models were significant in bivariate models for the majority the PSI subscales (α=0.05), and checked for multicollinearity. See the parameter estimates and statistics provided in the tables for details.

Table 5a demonstrates the changes over time in PSI subscales, moderated by the infants' health status (represented by the effect of *visit x CHD/healthy group* interaction term). PSI changes over time were significantly moderated by the infants' health status in the Reinforces Parent subscale (p=0.044), Mood subscale (p=0.010), Attachment (p=0.015), Role Restriction subscale (p=0.019), Parent Domain (p=0.026), Total Stress subscale (p=0.039), and Life Stress subscale (p=0.004). Table 5b and Figure 3 provide details of the moderating effects of the two groups over time on PSI. For the significant interaction effects, the opposite directions of the slope lines indicate that parents of subjects and controls differ in their PSI over time.

As indicated in Table 5b, for parents of healthy infants, Reinforces Parent scores insignificantly increased by 0.16 points every three months (p=0.128), and for parents of infants with CHD the scores insignificantly decreased by 0.18 points (p=0.128). Next, Mood subscale scores insignificantly decreased in the subjects group by 0.25 points (p=0.082), and insignificantly increased in the controls group by 0.23 points every three months (p=0.060). While Attachment scores of healthy infants' parents insignificantly increased by 0.09 points with each visit (p=0.386), the scores significantly decreased in the subjects group by 0.49 points every three months (p=0.000). As for the Role Restriction subscale scores, parents of healthy infants experienced insignificant stress

increase by 0.30 points (p=0.179), and parents of infants with CHD experienced insignificant stress decrease by 0.38 points in each visit (p=0.059). While the healthy infant parents' scores in the Parent Domain insignificantly increased by 0.46 points with each visit (p=0. 0.496), scores of parents of CHD infants significantly decreased by 1.90 points every three months (p=0.025). Further, Total Stress scores insignificantly increased by 1.741 points in the controls group (p=0.111), and insignificantly decreased by 1.84 points in the subjects group (p=0.192) every three months. Lastly, Life Stress scores of healthy infants' parents significantly decreased by 1.38 points (p=0.001) with each visit. On the other hand, scores of CHD infants' parents insignificantly increased by 0.19 points (p=0.532) every three months.

Whereas no other significant interaction effects were demonstrated on the rest of the PSI subscales, Table 5b also shows significant main effect of time (visit) on the Distractibility subscales, and the Child Domain. For parents of healthy infants, both Distractibility scores and Child Domain scores significantly increased with each visit, by 0.45 (p=0.023) and by 1.36 points (p=0.033), respectively.

Table 5c presents a sub-group analysis of the changes over time in PSI subscales in parents of infants with CHD. The relationship is moderated by the infants' post-op cardiac physiology (Single-ventricle vs. Biventricular functioning heart), and represented by the *visit x physiology* interaction term effect). In parents of infants with CHD, PSI changes over time were significantly moderated by the infants' cardiac physiology in the Distractibility (p=0.002), Mood (p=0.009), and the Child Domain (p=0.023) subscales.

Similarly to the previous analysis, the effects of "Visit" on PSI are individually displayed for SV/BV groups in Table 5d and in Figure 4.

For the significant interaction effects, the opposite directions of the slope lines indicate that parents of SV and BV infants differ in their PSI change over time. As indicated in Table 5d, Distractibility stress scores of SV infants' parents insignificantly decreased by 0.26 points every three months (p=0.476), while for parents of BV infants the scores significantly increased by 1.13 points (p=0.003) every three months. Mood scores of parents of SV infants significantly decreased in by 0.59 points every three months (p=0.026), whereas for parents of BV infants, the increase by 0.02 points with each visit was insignificant (p=0.908). Finally, stress scores in the Child Domain insignificantly decreased by 2.00 points in each visit for the SV group (p=0.188), and insignificantly increased by 1.17 points in the BV group (p=0.265).

Table 5d further demonstrate significant main effects of *Visit* on additional PSI subscales. Interestingly, both parents of SV and BV infants demonstrated highly significant decrease of 0.61 and 0.47 points (respectively) in stress over time on the Attachment subscale (p=0.004, and p=0.002; respectively). Additional significant PSI changes over time were experienced by parents of SV infants on the Role Restriction subscale, with a decrease of 0.61 points at each visit (p=0.043); Parent Domain, with a decrease of 2.53 points at each visit (p=0.043); and Total Stress subscale with a decrease of 4.51 points at each visit (p=0.031).

Aim 3. Aim 3 sought to examine associations between parenting stress and infant neurodevelopment in CHD and healthy infants. Tables 6a-6l summarize results from

multivariable regression models for MDI and PDI at six and twelve months. Each table represents a set of models, in which development (MDI and PDI) is regressed on a single PSI subscale at a single time point (three, six, nine, or twelve months). Within the controls group, all models are adjusted for infant length Z-scores at the measured PSI time point, and for gestational age; within the subjects group, all models are adjusted for infant length Z-scores, gestational age, and length of hospital stay (LOS). All covariates included in the multivariable models were significant at 6 or 12 months for either one or both outcomes (α=0.05), and checked for multicollinearity. See the parameter estimates and statistics provided in the tables for details.

Results for MDI at six months and PSI at three and six months are presented in Tables 6a and 6b, respectively. Table 6a demonstrates that for healthy infants, higher stress scores on the Spouse subscale at three months of age were associated with higher MDI scores, later at six months of age (p=0.048). For infants with CHD, higher stress scores on the Parental health subscale were associated with lower MDI scores at six months of age (p=0.034). The total variance (R^2) for MDI at six months accounted by this set of models ranged between 27-35% for the controls group, and between 40-48% for the subjects group. Table 6b shows no significant associations between PSI and MDI scores at six months of age for healthy infants. However, higher scores on the Parental Reinforcement (p=0.047), Isolation (p=0.047), and Depression (p=0.028) subscales on the PSI Parent Domain (overall p=0.044) at six months, were all significantly associated with lower MDI scores of infants with CHD at six months of age (R^2 =31-40%).

Results for PDI at six months and PSI at three, and six months are presented in Tables 6c and 6d, respectively. Table 6c demonstrates that higher Distractibility subscale scores at three months were associated with higher PDI scores in healthy infants at six months of age (p=0.036; R^2= 2-14%); and that higher Parental Reinforcement subscale scores at three months were associated with higher PDI scores in infants with CHD at six months of age (p= p=0.045; R^2= 21-32%). As for six months (Table 6d), higher Parental Reinforcement stress scores were associated with lower PDI scores in healthy infants (p= p=0.035; R^2= 4-19%). No significant associations were demonstrated at six months between PSI and PDI scores in healthy infants.

Further, Tables 6e- 6h present the multivariable models' results for MDI at twelve months and PSI at three, six, nine, and twelve months. No significant associations were demonstrated between PSI at three months and MDI at twelve months in healthy or CHD infants (see Table 6e). Higher stress scores at six months on the Adaptability (p=0.044), and Parental Reinforcement (p=0.002) in the Child Domain (overall p=0.030), and Parental Attachment (p=0.048) subscale were all associated with lower MDI scores at twelve months of age in the healthy group (R^2=18-41%). The subjects group demonstrated no significant associations between PSI at six months and MDI at twelve months (see Table 6f). Table 6g shows that higher stress on the Parental Health subscale at nine months was associated with higher MDI scores at twelve months of age in healthy infants (p=0. 002); and that higher Role Restriction stress at nine months was associated with higher MDI scores in infants with CHD at twelve months (p= p=0.035). R^2 ranged between 1-40% for the controls group, and between 44-53% for the subjects group.

Lastly, no significant associations between PSI and MDI were demonstrated at twelve months in the healthy group. However, higher PSI scores on the Acceptability (p=0.029), Competence (p=0.048), Isolation (p=0.015), Role Restriction (p=0.005), Parent Domain (overall p=0.024), and Total Stress (overall p=0.046), were significantly associated with lower MDI scores at twelve months of age (R^2=22-32%).

Similarly, Tables 6i- 6l present the multivariable models' results for PDI at twelve months and PSI at three, six, nine, and twelve months. No significant associations were demonstrated between PSI at three months and PDI at twelve months in healthy infants (see Table Xi). For infants with CHD, higher Role Restriction scores at three months were associated with higher PDI scores at twelve months (p=0.003; R^2=20-34%). No significant associations were found between PSI at three months and PDI at twelve months in healthy infants (Table 6j). As for the subjects group, while higher stress on the Mood subscale at six months was associated with higher PDI scores at twelve months (p=0.010), higher Role Restriction and Life Stress scores were both associated with lower PDI scores at twelve months (p=0.040 and 0.039, respectively). R^2 in this group ranged between 20-34%. Table 6k shows that higher stress on the Parental Health subscale at nine months was associated with higher MDI scores at twelve months of age in healthy infants (p=0. 040); and that higher Life Stress at nine months was associated with lower PDI scores in infants with CHD at twelve months (p= p=0.026). R^2 ranged between 2-18% for the controls group, and between 29-42% for the subjects group. Finally, no significant associations between PSI and PDI were demonstrated at twelve months in the healthy group (Table 6l). In the subjects group, higher PSI scores on the Distractibility

and Mood subscales were associated with higher PDI scores (p=0.010 and p=0.044, respectively); and higher Role Restriction stress was associated (p=0.005) with lower PDI scores at twelve months of age (R^2=18-27%).

Power Analysis

Power calculations for the current study were performed using PASS13 Power Calculation Software (PASS13, 2014). For aim 1, two-sample independent t-tests were used to determine the Minimal Detectable Differences (MDD) with 80% power (α=0.05), for all PSI subscales at each time point. The MDD is an estimate of the smallest statistically significant difference that can be detected when comparing two group means (Brock et al., 2015; Revicki et al., 2006). In our case, the MDD demonstrates the smallest significant difference we were able to detect when comparing means of parenting stress between CHD and healthy infants. For example, at three months of age, we were able to detect a significant difference in Total Stress mean scores of 18.3 or higher, given the estimated variances of the subjects and the controls groups (sd=±34.17 and ±38.04, respectively; see Table 7a). Additional MDD estimates for various PSI subscales by the various time points are presented in Table 7.

Post-hoc power analyses were performed for aims 2 and 3. For aim 2, two-sided Wilcoxon tests (Al-Sunduqchi, 1990; Machin, Campbell, Fayers, & Pinol, 1997) were performed to determine the power needed to detect a significant interaction Visit x CHD/Healthy effect for each PSI subscale (α=0.05). For example, a sample size of 123 achieves 76% power to detect an interaction effect of 3.69 on Total Stress subscale at the 0.05 level of significance. Similarly, a sample size of 123 achieves 93% power to detect

an interaction effect of 1.48 on Life Stress subscale at the 0.05 level of significance. See Table 7b for detailed analysis.

For Aim 3, F-Tests (Cohen, 1988) were used to estimate the minimal detectable R^2 attributed to each PSI subscale in explaining the variation for the developmental outcomes (MDI and PDI at three and six months), with 80% power and a significance level of 0.05. Control variables included length Z-scores and gestational age for the healthy infants, and length Z-scores, gestational age, and LOS for the CHD infants. For example, a sample of 37 healthy infants achieves 80% power to detect an R^2 of 0.14 attributed to Total Stress subscale at three months, in predicting MDI at six months. This R^2 is added to the R^2 of 0.24 explained by infant length Z-scores, and gestational age, and sums up to a total R^2 of 0.38. Additional examples for R^2 power calculations for Total Stress subscales on the developmental outcomes at the various time points are presented in Table 7c.

CHAPTER 5- DISCUSSION

Introduction

This study sought to explore parenting stress in parents of infants with CHD over the first year of life, and compare it to the stress in parents of healthy controls. It also examined the associations between parenting stress and neurodevelopmental outcomes over the first year of life in infants with CHD and healthy infants. This final chapter will summarize the study findings and will discuss their meaning in depth, referring to the existing literature. Further specification of the theoretical and practical implications of the findings will follow. The study's limitations will be identified, concluding with suggestions for future research.

Discussion and Summary of Principal Findings

Parenting Stress in Parents of Infants with CHD and of Healthy Infants

To the best of my knowledge, this is the first study to investigate the levels and changes of parenting stress over time in the CHD population during infancy, and compare them to healthy controls. The objective of Aim 1 was to describe and compare levels of parenting stress between parents of infants with CHD and parents of healthy controls at three, six, nine, and twelve months of infant's life. Findings demonstrate that at three months of age, parents of infants with CHD experienced significantly higher parenting stress than parents of healthy controls on the Demandingness, Mood, Child Domain, and Competence subscales. The stress on the Demandingness subscale remained significantly higher in parents of infants with CHD compared to parents of healthy infants, throughout

the first year of infants' life. Additionally, parents of infants with CHD experienced significantly higher stress on the Life Stress subscale than parents of healthy infants at twelve months of age.

The parenting stress literature over the years has reported higher parenting stress among parents of children with illnesses compared to parents of healthy (Maas-van et al., 2013; Rabineau et al., 2008). Specifically, in parents of children with CHD, higher parenting stress has been documented by many (Darke & Goldberg, 1994; Goldberg et al., 1991; Rimmerman & Stanger, 2001; Torowicz et al., 2010). In some cases, the parenting stress experienced by parents of infants with CHD was even higher than those of infants with other chronic healthcare needs such as Down's Syndrome, Autism, and more (Goldberg et al.,1990; Mullen et al., 2014).

The main differences in the early stress at three months appear to be on the Demandingness, and Mood subscales within the Child Domain, and on the Competence subscale within the Parent Domain. According to Abidin (1995), elevated scores on the Child Domain usually result from child's qualities that make it difficult for parents to fulfill their parenting roles (See Table 8 for subscales' short descriptions). Specifically, elevated stress scores on the Demandingness subscale suggest that the parents perceive the child as very dependent, or placing many demands on them; and elevated scores on the Mood subscale suggest that the child displays dysfunctional affective behaviors such as excessive crying, or seem unhappy in general. Many other studies similarly found increased stress on the demandingness subscale, and on the Child Domain in infants and children with CHD (Brosig et al., 2007; Dudek-Shriber, 2004; Goldberg et al.,1990;

Uzark & Jones, 2003). These findings align well with the CHD literature, in which some temperamental traits of pediatric CHD population have been described to be particularly challenging for parents. Infants with complex CHD have been described as more irritable, intense, difficult to sooth, and having more negative mood than healthy controls (Carey et al., 2002; Darke & Goldberg, 1994; Marino & Lipshitz, 1991). Such temperamental traits are often attributed to the birth and hospitalization experiences of premature infants, infants with CHD, and other groups with congenital defects. These experiences often involve hypoxemia, multiple medications, feeding difficulties, and growth failure (Carey et al., 2002; Hughes, Shults, McGrath, & Medoff-Cooper, 2002; Wernovsky, 2006), leading eventually to increased burden of care for parents (Farley et al., 2007; Torowicz et al., 2010).

The finding of higher stress on the Competence subscale in parents of CHD infants compared to healthy controls, is supported by the literature as well, especially in complex CHD groups (Brosig et al., 2007; Goldberg et al., 1990). Elevated stress scores within the Parent Domain, and specifically on the Competence subscale, suggest that the sources of stress emerge from the parent-child system, and may relate to parental and/or child's characteristics, such as young parental age, gaps between the parental expectations and reality, and major mental or physical disability of the child (Abidin, 1995). Indeed, parents of CHD infants across studies reported experiencing difficulties in balancing parental-role functions (Dudek-Shriber, 2004; Hayes & Knox, 1984; Farley et al., 2007; Sarajuuri et al., 2012; Young-Seideman et al., 1997), difficulties in setting limits, and disciplining their child (Uzark & Jones, 2003). A possible explanation might

connect this finding to the previous findings of higher stress on the Child Domain. Secco & Moffatt (2003) suggested that 'difficult' temperaments or child's behavior can present a greater challenge for parents, which in turn, might contribute to parenting sense of incompetence. I may also be that parents feel inadequate because they are often not in control of their infant's health issues and cardiac complications.

Parents of infants with CHD also experienced higher stress on the Life Stress subscale later at twelve months. Elevated Life Stress scores indicate that parents have to deal with stressful situational circumstances outside the parent-child system, and these usually intensify the parenting stress. The questions regarding life stress in the PSI refer to events occurring during the last 12 months, therefore stressful evens any time prior to that time point could be influential on later parenting stress (Abidin, 1995). This stress may be due to the uncertainty of infant outcomes, or related to the ongoing demands on the parents such as monitoring multiple medications, ER visits, and hospitalizations during the first year. Mullen et al. (2014) reported close correlations between the quality of life and parenting stress in parents of CHD children, even after controlling for disease severity. Similarly, parenting stress was more closely related to family resources than to the child's health status in Phipps and Drotar's (1990) study. Rimmerman and Stanger (2001) reported higher parenting stress in mothers of children with congenital conditions (CHD amongst them) than in healthy controls, and negatively associated those stress levels to the mothers' marital quality.

The interpretation of the PSI scores is based on percentile scores which were derived from the frequency distribution of a normative sample (by age) (Abidin, 1995).

In our sample, parents of infants with CHD demonstrated significantly higher parenting stress than controls. However, subjects' stress levels are considered to be only moderately high, with Demandingness scores ranging within the 60th-65th percentile compared to the 25th percentile in the healthy group; Mood scores in the 60th percentile compared to the 35th percentile in the healthy group; and Life Stress in the 70th percentile compared to the 55th percentile in controls. Surprisingly, the Child Domain scores were only in the 45th percentile compared to the 30th percentile in controls; and the Competence scores in the 25th compared to the 15th in the healthy. This trend of lower scores than expected across both groups (healthy controls should correspond to the 50th percentile) is surprising and might result according to Abidin (1995), from several factors, amongst them are defensive, or what parents believe is socially desirable responding to the questions, or disengagement in the parental role.

Parenting Stress Over Time

The objective of Aim 2 was to identify changes in stress levels in parents of CHD infants over the first year of life, and compare them to those of healthy controls. Findings indicate that the Total Stress change over time (i.e. increase/decrease) significantly differed in parents of subjects and controls, both on the Child Domain (Reinforces Parent and Mood), and on the Parent Domain (Attachment and Role Restriction). The Life Stress change over time differed between the groups as well. These differences were mostly indicated by opposite directions in the stress change over time. While stress of parents in the subjects group has decreased with time, parents of controls tended to experience increase in stress with time. Nonetheless, a separate examination of the stress

for each group revealed significant change over time in only several subscales. For parents of infants with CHD, stress has decreased on the Attachment subscale within the Parent Domain; and for parents of healthy infants, stress has increased on the Distractibility subscale within the Child Domain. Also, their Life Stress has decreased over time. Deeper examination of the subjects group by their post-operative physiology revealed that the two sub-groups significantly differed in their stress change over time on the Distractibility and Mood subscales within the Child Domain. As for the sub-group individual analysis, parental stress of BV infants has increased on the Distractibility subscale, and decreased on the Attachment subscale. In the SV infants group, the Total Stress resulting from the Attachment and Role Restriction subscales within the Parent Domain, and Mood within the Child Domain, has significantly decreased over time.

In the current study the parenting stress in the subjects group has decreased over time, whereas in the healthy group, in large part, stress has increased over time. According to Abidin (1995), there is a general expectation of the Total Stress to decrease as the child ages from 1-3 years. The change in parenting stress within the first year, however, is not discussed, nor does the CHD pediatric population. Studies reported mix findings with regard to what happens to parenting stress over time across pediatric populations. Both Dyson (1993) and Crnic et al. (2005) found stable parenting stress levels in healthy preschool children over time. Stable stress levels have also been reported in parents of children with disabilities, and children with ASD (Ben-Sasson et al., 2013; Dyson, 1993; Peters-Scheffer et al., 2012). Rivard et al. (2014) reported on increases in parenting stress over time in children with ASD, while others reported on

decreases in parenting stress in children with ASD (Mak et al., 2007; Osborne & Reed, 2009) and children with cancer (Fedele et al., 2011; Tsai et al., 2013). The stress in these studies has been associated with the severity of the illness; and the decreases over time have been mostly explained by the parental adjustment to the situation and/or by the reduction/diminishing of treatments over time (Fedele et al., 2011; Tsai et al., 2013). The increase in stress in parents of healthy infants may be explained by the fact that mothers of these infants often return to work after several months postpartum, which may contribute to the stress they feel (Nichols & Roux, 2004; Nyström & Öhrling, 2004).

With regard to CHD populations, Uzark and Jones (2003) showed that stress increased with age in parents of children aging between 2-12 years. These cross-sectional associations were explained by increasing challenges to discipline and setting limits as children age. It is probable that the infancy period possesses different parenting challenges or concerns, which results in different stress characteristics than in parents of older children. Such challenges might be attributed to changing patterns in child's temperament (Marino & Lipshitz, 1991; Schraeder & Medoff-Cooper, 1983; Spungen & Farran, 1986), developmental status (Osborne & Reed, 2009; Robson, 1997), and other illness-related factors. For example, prenatal diagnosis is known to be one of the stress contributing factors in mothers of CHD infants (Pintoa et al., 2016; Rychik et al., 2013; Wei, et al., 2016). The neonatal heart surgery, the stay in the CICU, and the following hospital discharge to home, are other good examples for critical stressful periods (Hartman & Medoff-Cooper, 2012; Harvey, Kovalesky, Woods, & Loan, 2013; Solberg, et al., 2012; Wei et al., 2016). Following discharge to home there is a stressful period for

parents who have to deal with feeding difficulties, learn how to manage their child's condition, and face the threat on their child's life (Harvey et al., 2013). Gaskin et al. (2016) showed decrease in signs of PTSD in parents of infants who underwent cardiac surgery, as their confidence increased.

These reports may also provide explanation to the finding of decreasing stress on the Attachment and Reinforces Parent subscales in the subjects group. Stress on the Attachment scale suggests either that the parent does not feel emotional closeness to the child, nor that the parent is able to observe/understand the child's needs. Attachment related Stress is often reflected in cold parent-child interaction. Subsequently, stress on the Reinforces Parent subscale indicates that the parent- child interaction fails to produce good feelings by the parents about themselves, that the parent might feel rejected by the child, or that the parent-child bond is threatened (Abidin, 1995). Others have indicated insecure attachment bonds, and weak infant-mother relationships experienced by mothers and their infants with congenital and chronic illnesses including CHD, in comparison with healthy peers (Goldberg et al., 1991; Mäntymaa, Puura, Luoma, Salmelin, & Tamminen, 2006). The general explanations suggest that the difficulties to engage in mother-infant interactions stem either from the mother or from the infant. For example, mothers of seriously or recurrently ill infants might have psychological and physical barriers in the bonding and attachment processes due to the long hospitalizations in the ICU environment, and/or the uncertainty in the infant's survival (Board & Ryan-Wenger, 2000; Young Seideman et al., 1997). Additionally, complex CHD infants often lose attention and quickly withdraw during interactions, challenging their care providers to

maintain those interactions (Gardner, Freeman, Black, & Angelini, 1996). The fact that this may improve with time (Gardner et al., 1996) aligns with our findings of decreasing stress on the two subscales in the subjects group over time.

Along the continuum of outcomes, we also saw decrease in stress over time in mothers of SV infants compared to BV infants, especially on the Mood and Role Restriction subscales. Stress on the Role restriction stress derives from parental frustrations that their self-identity and personal freedom are restrained because of their parental duties. The temperamental changes in CHD infants over the first year of life are evident, especially in those with complex conditions (Wernovsky, 2006). As stated before, the nature of the complex defects imposes biological strains and serious medical interventions that have adverse effects on infant's behavior (Hughes et al., 2002), and on the burden of care (Torowitcz et al., 2010). It is reasonable to believe that stress reduces as these surgical effects and the burden of care lessen with time, and/or as parents learn how to cope with the illness.

Finally, with regard to the clinical interpretation of the stress change over time, whereas the stress change significantly differed between the groups, it did not reach clinical levels within the significant subscales. The stress in the sample ranged between the 25^{th} - 50^{th} percentiles for the different subscales, which are moderately low levels. The only exception was the Life Stress of healthy controls, which dropped from the 75^{th} to the 55^{th} percentile over time.

Parenting Stress and Infant Neurodevelopment

Aim 3 sought to examine associations between parenting stress at three, six, nine, and twelve months, and infant neurodevelopment at six and twelve months in CHD and healthy infants. Consistent with previous reports, the PDI and MDI scores both are below standardized means for the subjects in our sample, with MDI being less severely affected (Fuller et al., 2009; Long, Galea, Eldridge, & Harris, 2012; Mussatto et al., 2014). Low MDI scores represent delays in higher-order mental processing such as language acquisition, reasoning, memory, and integration of these processes (Aylward, 1997). In young infants MDI assesses visual and auditory habituation, and problem-solving skills such as object permanence, perspective taking, and following multistep directions. Low PDI scores indicate possible delays in gross and fine motor skills. In younger infants the PDI mainly assesses movement symmetry, and antigravity movement (thrusts arms and legs in play, lifts head, balances head). Delays in MDI and PDI can signal neurological impairment, oral motor impairment, general cognitive delay, or environmental deprivation (Bayley, 1993). In infants with CHD neurodevelopmental delays are usually predicted by prematurity, longer postoperative length of stay (Fuller et al., 2009), infant's length more than weight (Nydegger & Bines, 2006; Ravashinkar et al., 2013), and additional factors (Wernovsky, 2006). After adjusting for such factors, our analysis reveals several interesting relationships between parenting stress and infant neurodevelopment.

In the current study early parenting stress was associated with later developmental outcomes in both healthy and CHD infants. For healthy infants, higher stress on the Adaptability and on the Parental Reinforcement subscales within the Child Domain, and

on Parental Attachment at six months was associated with lower MDI at twelve months. Stress on the Adaptability subscale often indicates that the parenting tasks are being more difficult due to child's characteristics that make him/her unable to adjust to changes in physical or social environments. Such characteristics include overreaction to changes and stimulation, difficulties in soothing once upset, and more (Abidin, 1995). Those parents report being extremely frustrated in their attempts to develop a relationship with their child. According to Abidin (1995), elevated stress on this subscale, in combination with elevated stress on the Reinforces Parent and the Attachment subscales strongly indicate a weak parent-child relationship. This assumption aligns well with the above findings, suggesting a link between early stress, and later delayed mental development, through the mother-infant relationship.

For infants with CHD, higher Role Restriction stress and higher Life Stress at six months was associated with lower PDI scores at twelve months. These associations were also existing at twelve months. It is agreed that parents have the greatest opportunity in promoting their child's motor development (Mahoney & Perales, 2006; Noel, Peterson, & Jesso, 2008). It is possible that parents of infants with CHD have to deal with daily illness-related circumstances, which prevent them from investing time in the motor stimulation of their child. Mothers who reported high stress provided low levels of physical stimulation regardless of child temperament (Noel et al., 2008).

Findings also demonstrate several cross-sectional relationships between parenting stress and infant neurodevelopment. For infants with CHD, higher Parental Reinforcement stress within the Child Domain, and higher stress on the Isolation and

Depression subscales within the Parent Domain were associated with lower MDI at six months. At 12 months, higher Total Stress resulting from the Acceptability within the Child Domain, and from the Competence, Isolation, and Role Restriction within the Parent Domain, was associated with lower MDI. Stress on the Acceptability subscale usually signals that the child is not as attractive, intelligent, or pleasant as the parent had expected or had hoped, and might consequently result in poor attachment and/or rejection from the parent (Abidin, 1995). For healthy infants, higher Parental Reinforcement stress was associated with lower PDI at six months. It is unreasonable that higher stress at a certain time point has immediate effect on infant's development, rather it is more likely to assume the other way around. Numerous studies reported on a reciprocal/bidirectional model of the effects of parenting stress on child's behavior problems (often are predictors of developmental delays; see in Baker, et al., 2002), and vice versa (Baker et al., 2002; Mackler, et al., 2015; Neece, Green, & Baker 2012; Zaidman-Zait et al., 2014). Therefore, it is reasonable to believe that infants with delayed development may not meet their parents' expectations, and not provide enough reinforcement to their mothers, which raises their stress levels in turn. These findings were much more dominant in the CHD group, which possesses high incidence of developmental delays (Wernovsky, 2006). Possibly the developmental delays lead to parental feelings of incompetence and restrictions in the parental role, increasing parental feelings of depression and isolation.

Finally, some findings indicate unexpected associations in terms of their directionality. In the healthy group for instance, stress sourcing from parental health issues at six and nine months was *positively* associated with MDI at 12 months. Early

stress on the spouse subscale at three months predicted higher MDI at six months. High stress on this subscale indicates lack of emotional and active support of the other parent in the area of child management. In some instances, this is related to an overly strict sex-role definition on the part of the father that child's care is women's work (Abidin, 1995). These findings are surprising and are not fully understood. It is possible that a certain SES-related mechanisms and family functioning styles mediate/moderate these relationships between parental characteristics and infant's development. A literature search for a possible explanation revealed mixed results. Challahan (1989) raised the possibility that low SES parents, who report high levels of parenting stress, are likely to seek more medical care. Bakel and Riksen-Walraven (2002) tested a process model in which factors such as parental intelligence, ego-resiliency, education, partner support, and infant social fearfulness were found to explain significant portions of variance in the parental behavior, which in turn, was linked to the infants' attachment security and cognitive development. On the other hand, a meta-analysis of 68 studies examining possible moderators between marital relations to parent-child relations revealed no existent moderators, suggesting that this link is more stable than previously thought (Erel & Burman, 1995). The chronic pediatric literature also emphasizes the importance of family management and functioning to family and child outcomes. Knafl and colleagues (2013) showed lower degree of behavior problems in children raised by Family-Focused caregivers compared to Condition-Focused caregivers. Hocking et al. (2011) have linked family functioning variables to neurocognitive outcomes in brain tumor survivors. In the

current study, the limited SES data and family functioning parameters makes it is impossible to draw any conclusions regarding their impact on the examined relationships.

Such 'opposite-directional' relationships were similarly found in infants with CHD as well. Greater stress on the Parental Reinforcement subscale at three months was associated with higher PDI at six months. Role Restriction stress at nine months was positively associated with later MDI at 12 months; and at three months with PDI at 12 months. Higher stress on the Mood subscale at six months, and on the Distractibility subscale at 12 months was associated with higher PDI at 12 months. Similar finding was also found in the healthy group, in which higher stress on the Distractibility subscale at three months predicted higher PDI at six months of age. These associations might be explained by the interrelations between infantile and parental behaviors. It is possible that more active, moody, or distractible infants, solicit more parental response, which eventually promotes their development. It might also be that parents who more actively stimulate their infants, tend to feel more stressed and restrained by their parental role because they invest more time and efforts stimulating their infants. Susman-Stillma et al. (1996) found that maternal sensitivity mediated the relationship between infant irritable temperaments and their secure attachment. Crockenberg & Acredolo (1983) found some aspects of infant temperament to be associated with antecedent and concurrent mother behavior. Numerous studies have also demonstrated a link between mothers' mental state to infant development through a path of maternal responsiveness (Beck, 1998; Milgrom, Westley, & Gemmill, 2004; Pearson et al., 2012). Gandour (1989) showed that

parental stimulation differentially affects development depending on child's activity level.

Although the literature provides us with some potential explanations, it is more reasonable to assume that such opposite associations derive from a type 1 error. A type I error (or a "false positive") is the incorrect rejection of a true null hypothesis, which leads one to conclude that a supposed effect or relationship exists when in fact it doesn't. All hypotheses testing have the probability of making type I errors. This probability however, increases tremendously in exploratory studies, when researchers generate simultaneously multiple P-values, a situation known as the Multiple Testing Problem (Neyman & Pearson, 1933; Proschan & Waclawiw, 2000; Wright, 1992). The majority of the findings in the current study demonstrate negative associations between parenting stress and infant neurodevelopment, especially on the child's temperamental characteristics and parental attachment and competence domains, findings which are also strongly supported by previous research. Hence, such results resemble real, rather than being type 1 errors. On the other hand, we need to keep in mind that some relationships might derive from the Multiple Testing Problem, especially relationships between scattered PSI subscales and development.

Implications for Research, Practice and Policy

The current study utilized existing data to tackle novel research questions in a population of critically ill children and their parents. The innovative longitudinal design allowed us tracking and comparing parenting stress changes over time, and by that shedding some light on illness or specific sensitive infancy periods. To the best of my

knowledge, this was also the first study to examine the effect of parenting stress on infants' neurodevelopment in pediatric CHD population, which struggles with increased incidence of developmental delays. To date, the contributing factors to the delays in this pediatric population were mostly attributed to biology and clinical parameters. This study introduces psychosocial/familial aspects as additional contributors to infant development. Throughout the study we witness how important mother-infant attachment issues are, by being stress provokers, and by their relations to adverse developmental outcomes. Such findings have also been emphasized in the infant-maternal biobehavioral and mental health research, which suggests moving forward from linear models of cause and effect, towards multidimensional biopsychosocial models in the attempt to explain developmental psychopathology (Burgess, Marshall, Rubin, & Fox, 2003; Simmons, Goldberg, Washington, Fischer-Fay, & Maclusky, 1995; Wachs, 2009S; Zeanah, Boris, & Larrieu, 1997). Zeanah et al. (1997) suggest that infant development is best appreciated within the context of caregiving relationships, which may mediate the effects of both intrinsic and extrinsic risk conditions. The mediating/moderating role of parenting stress should be examined in such models as a potential key indicator of dysfunctional interactions in the parent-child relationships (Gordon & Hinshaw, 2015).

Findings indicated that parents of infants with CHD generally experience higher parenting stress levels than parents of healthy infants, and that their stress reduces with time. It might be concluded that the stress evokes around child temperamental characteristics, and also probably around illness-related burden. Findings align with the literature which highlight the post-operational and hospital discharge periods as sensitive

stressful periods for parents, as they begin to adjust to their infant's illness and start learning how to cope with it in the home settings (Franck et al., 2010; Hartman & Medoff-Cooper, 2012). Socio-familial factors such as healthy partner relationships, cohesiveness, and adaptive parental coping mechanisms are necessary for successful parental adaptation to CHD in their child (Rychik et al., 2013; Sira, Desai, & Hannon, 2014). Families who have fewer psychosocial resources and lower levels of support may be at risk of higher psychological distress and lower well-being over time (Jackson, Frydenberg, Liang, Higgins, & Murphy, 2015). The experiences, needs and coping strategies in families of children with CHD are multi-faceted, and may even change over time (Gray, 2006; Rempel, Ravindran, Rogers, & Magill-Evans, 2013). Gray (2006) reported that early coping mechanisms in parents of children with autism include reliance on service providers and family support, whereas with time these change to more emotion-focused coping strategies (e.g. religious faith). Similarly, Tak and McCubbin (2002) found social support to be important predictor of family coping following the diagnosis of CHD. Early intervention should aim to promote parental adaptive coping and productive parenting practices in this population (Jackson et al., 2015).

A recent systematic review of the literature indicated that the most effective interventions in reducing parenting stress were those who strived to change families' illness perceptions and coping mechanisms (Golfenshtein, Srulovici, & Deatrick, In Press). Such interventions strengthen the connections of coping and adjustment models in families who experience these extraordinary stressors (Lazarus & Folkman, 1984;

McCubbin & McCubbin, 1993; Patterson, 1988; Wright & Bell, 2009). Research has also demonstrated great effectiveness of interventions that included multiple family members, and focused on specific family needs (Conners, Edwards, & Grant, 2007; Pouretemad, Khooshabi, Roshanbin, & Jadidi, 2009; Verreault, Verret, Massé, Lageix, & Guay, 2011). Under Abidin's Model, major life events are perceived as separate sources of stress for parents. Although CHD is a stressful life event by all means, findings indicate that the parenting stress experienced by parents of infants with CHD is integrated into almost every aspect of family life. It frequently emerges from multiple factors, and involves the various members of a family. Therefore, interventions that reduce parenting stress need not exclusively target parenting processes directly (Deater-Deckard, 2004).

The Illness Belief Model shifts the illness focus from the individual (the child) to include family members and healthcare provides, by emphasizing the role of the illness beliefs held by individuals, families, and healthcare providers, in shaping the illness experience and the healing process (Bell & Wright, 2011; Bell & Wright, 2015). Family systems level interventions may have the greatest leverage for families dealing with illness related distress, by focusing on the familial strengths, providing social support and sharing information, and eventually enhancing familial competence to cope with the illness (Atkin & Ahmad, 2000; Bell, 2013; Chesla, 2010; Dunst & Trivette, 2009; Wacharasin et al., 2015; West, Bell, Woodgate, & Moules, 2015). Nevertheless, the optimal length and timing to intervene are not agreed upon. It has been argued that parenting stress cannot be permanently reduced in brief interventions (Sung, Gi-Do, & Park 2012). The lasting effects of parenting stress reduction interventions usually range

between few weeks and several months (Golfenshtein et al., In Press). In order to prolong the intervention effects, studies suggest establishing a comprehensive and long-term system of family care (Abedin & Molaie, 2010; Sung et al., 2012). Such family care system naturally resembles the Family Systems Nursing, which is the clinical application of the Illness Belief Model (Wright & Bell, 2009; Wright & Leahey, 2013).

The dialog regarding the shift of nursing care towards a family systems paradigm has intensified in recent years (Bell, 2009; Duhamel, Dupuis, Turcotte, Martinez, & Goudreau, 2015; LeGrow & Rossen, 2005). It is especially relevant to parenting stress, because of its psychosocial context to healthcare concerns (Duhamel et al., 2015). Family systems models with foci related to parenting stress can be incorporated into healthcare education and organizational policies (Bell, 2013). In order that to happen, healthcare students and providers can be taught how to recognize the various stressors affecting parents in different periods, by mastering therapeutic conversations with families (Bell, 2013). Family systems research demonstrated that educational interventions (e.g. video recording of therapeutic conversations to students, live supervision, and family assessment tools) helped nurses to develop confident in their abilities to establish solid relationships with patients (Bell, 2014; Konradsdottir & Svavarsdottir, 2011; Tapp, Moules, Bell, &Wright, 1997). Such skilled nurses may act to identify family competencies and strengths, and tailor stress-reduction interventions through this complex knowledge (Bell, 2014; Houger, Limacher, & Wright, 2006; Streisand, Rodrigue, Houck, Graham-Pole, & Berlant, 2000).

Study Limitations and Directions for Future Research

Sample-Related Limitations

The study possesses several design problems and limitations inherent to the secondary data analysis. The convenience sample was recruited from a single institute within a specific setting, a fact that might have led to homogeneous sample, limiting the external validity of the study and the generalizability of results. Additionally, the paucity of demographic and SES characteristics made it less comparable to the general population, and limited the ability to control for other potential confounders in the analysis. Similarly, the lack of a general measure of family functioning limited the ability to draw a full picture of the phenomenon. On the same note, the study's sample mostly included mothers. Mothers are often the primary caregivers in infancy, and therefore, most prior parenting stress research was conducted with samples of mothers (Carey et al., 2002; Phipps & Drotar, 1990; Rimmerman & Stanger, 2001). Studies who examined both parents, however, showed different characteristics of parenting stress in mothers and fathers (Dudek-Shriber, 2004; Goldberg et al., 1990; Sarajuuri et al., 2012). The family-systems perspective advocates for research based on multiple family members in order to receive a comprehensive picture of the phenomenon (Bell, 2009; Chesla, 2010).

The limited sample size has also prevented us from applying methods adjusting for multiplicity (i.e. the Multiple Testing Problem). Studies testing multiple hypotheses typically apply procedures adjusting the alpha levels for the multiple tests performed, to maintain the type 1 error rate at or below the specified level (5%). Such strategies include, among others, the Bonferroni, the Holm's, and Hierarchical order closed test

procedures (Proschan & Waclawiw, 2000; Wright, 1992). All of these strategies, however, require a larger sample size than was available in the current study. Therefore, it is inevitable to find many type 1 errors within the multiple relationships assessed in the current study. Nevertheless, it is essential to discuss the clinical significance along with the statistical significance, and interpret the results while keeping in mind the grater picture. Most of the Findings similarly suggest that stress evoked by child's temperamental characteristics, and by parental sense of attachment and competence, is related to adverse developmental outcomes. These findings are also supported by previous research.

Analysis-Related Limitations

Missing data and dropout are common problems in longitudinal designs and secondary analyses (Hedeker & Gibbons, 1997). They were both accounted for in the analysis by the mixed modeling approach, and by the comparison to an imputed dataset, which had demonstrated no missing data related bias in our results. However, the abundance of the missing data limited the power for the current study by decreasing the sample size. Our post-hoc power analyses indicates that the power for aim 2 ranged widely from 5-93%, demonstrating small to moderate effect sizes for the different PSI subscales. For Aim 3, our sample size allowed us to detect a minimal R2 ranging between .09-0.23 for the various PSI subscales, which were not small enough (when added to the variance of the other covariates in the models) to detect a significant effect for all cases. The reported effect size estimates and variation for parenting stress in the developmental research field are scant. Nevertheless, other studies typically explain between 30-40% of

the developmental outcomes variation in their models, when including clinical and biological covariates (Karsdorp, Everaerd, Kindt, & Mulder, 2007; Medoff-Cooper et al., 2015). This implies that child development is complex and that the phenomenon is difficult to characterize in unidimensional models. The missing data also limited our ability to perform additional analyses for Bayley's Behavior Rating Scale (BRS), as initially proposed. The BRS, which assess the child's emotional, behavioral, social, and environmental orientation, could perhaps be the most predicted by parenting stress, as its measured developmental aspects have been reported to be directly related to maternal attachment and mother-infant relationship (Brown, Olson, & Croninger, 2010; Skovgaard, Houmann, Landorph, & Christiansen, 2004). Future research should account for the socio-emotional development of infants and children when discussing the effects of dysfunctional parenting.

In the attempt to track parenting stress over time in the current study, 'time' was used as a continuous measure. This provided us with a linear result of increase or decrease in stress over time. Non-linear analysis or categorical use of the 'time' variable might have provided us with more detailed information, possibly demonstrating peak stressful periods in infancy. Previous research has identified that the prenatal diagnosis, days and weeks around the surgery, and the early weeks at home are especially stressful for parents of infants with CHD (Franck et al., 2010; Hartman & Medoff-Cooper, 2012; Rychik et al., 2013). This population might benefit from longer assessment through infancy and beyond, involving qualitative evaluations of participants' experiences.

Finally, the data were partially collected via self-reporting questionnaires, involving a potential risk of social desirability bias (i.e. responses are idealized to conform to socially accepted norms). However, most of the conventional research in this field utilizes self-reporting methods, as parenting stress is mostly subjective measure of perceptions and attitudes. Lately, studies tend also measure physiological stress as a confirmative measure to the psychological distress (Harmon, Hibel, Rumyantseva, & Granger, 2007).

Measure-Related Limitations

Parenting stress was measured in the current study by the PSI, which is a reliable general parenting stress measure, validated in multiple populations, including the CHD population. Nevertheless, recent review of the stressors across pediatric conditions demonstrated that sources of parenting stress vary according to specific illness characteristics and their implications for families. Moreover, the review findings suggested that illness-related factors, which are currently not included in Abidin's model also have significant impact on parenting stress (Golfenshtein, Srulovici, & Medoff-Cooper, 2016). More illness-specific and/or illness-related parenting stress measures exist (Chan & Sigafoos, 2001; Streisand, Braniecki, Tercyak, & Kazak, 2001). Utilizing such measures may provide information beyond the obtained from the general measure (Streisand et al., 2003) but would hamper across condition comparisons.

Infant neurodevelopment was measured via the BSID-II, which has been in standard use at the time the parent study was conducted (Anderson, De Luca, Hutchinson, Roberts, & Doyle, 2010). Since then, the BSID-II has been revised to a third edition

(BSID-III; Bayley, 2005). Nevertheless, the use of the BSID-III has only been reported in scant in the literature, and only handful of studies have been published in which the Bayley-III was used to assess infants or toddlers with congenital heart disease (Acton et al., 2011; Long et al., 2012). Long and colleagues (2012) showed that infants with CHD had a lower percentage of developmental delays on the Bayley-III as compared to similar samples of infants with CHD who were tested on the BSID-II. They indicated that the Bayley-III may underestimate developmental delays, and recommend caution in interpreting Bayley-III scores for high-risk children.

Conclusions

The current study tackled novel research questions in a population of critically ill children and their parents, by tracking parenting stress over time, and examining the effect of parenting stress on infants' neurodevelopment. To date, the contributing factors to the delays in the CHD pediatric population were mostly attributed to biology and clinical parameters. This study introduces psychosocial/familial aspects as additional contributors to infant development, through a route of mother-infant relationship. Nursing practice should utilize Family Systems interventions in early stressful periods, which may be risky for parents of infants with CHD.

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Table 1

Study Variables and Measurements

Study Variables	Definition of term	Instrument (source) Subscales	Operational definition Items (score range)	Data Collection
1. Parenting stress	The psychological distress parents experience resulting from the parenting role.	PSI- Long Form* (Abidin, 1995)	Continuous	Self-reported by parents during 3,6,9, 12mo visits.
		Child Domain (CD) subscales	47 5-point Likert style scale items, sum of 6 subscales (47-235)	
		Parent Domain (PD) subscales	54 5-point Likert style scale items, sum of 7 subscales (54-270) Sum of CD & PD scores (101-505)	
		Total Stress (TS) subscale	19 items (0-81)	
		Life Stress (LS) subscale	19 items (0-81)	
2. Infant Neuro-development	The biological, psychological and emotional changes which determine the child's socio-emotional, and cognitive capacities, occurring in human beings in the process of maturation between birth and the end of adolescence.	BSID-II+ (Bayley, 1993)	Continuous	Assessed by developmental psychologist during 6 and 12 mo visits.
		Mental Scale	20-36 items designated by age creating mental development index (MDI; 50-150)	
		Motor Scale	14-21 items designated by age creating the psychomotor development index (PDI; 50-150)	
		Behavior Rating Scale (BRS)	30 items forming 4 standardized categories (Emotional, Behavioral, Social, Environmental orientation Percentiles)	

3. Post-operative Cardiac Physiology	Infant's cardiac functionality in terms of functioning ventricles after surgery, according to the defect type and surgery outcome.	Postoperative echocardiograms in medical records	Categorical Single ventricle (SV) Bi-ventricle (BV) Normal (healthy controls)	Assessed by pediatric cardiologist at enrollment.
4. Risk for In-Hospital Mortality+	The complexity and the primary post-surgical mortality risks for the cardiac defects and the surgical procedures performed in CHD patients.	RACHS-1 ** (Jenkins et al., 2002) Standardized risk categories	Categorical Ranging from 1-6 (lowest to high risk)	Assessed by pediatric cardiologist at enrollment.
5. Length of Stay	Number of days the infant stayed in the hospital after surgery.	Medical records	Continuous (Stay in days)	Recorded by research staff at discharge.
6. Feeding Mode	Infant's enteral way of feeding.	Medical records Oral feeding mode Device-assisted feeding mode	Dichotomous Breast and/or bottle Device-exclusive feeding; or breast/bottle feeding, supplemented by device (nasogastric, gastric, jujunal tube)	Recorded by research staff at discharge, and at 3mo visit.
7. Infant Growth	Infant's physical development as determined by measures of body proportions (anthropometry).	Anthropometrics measured via: Infant digital scale for Weight (Scaletronix, White Plains, NY)	Continuous Raw anthropometrics transformed into the WHO growth standardized z-scores (mean =0, SD=1).	Measured by trained research staff at 3,6,9,12 mo visits.

		Infant length board for Length (Holtain, Crymych, UK) Fiberglass tape for HC ŧ (McCoy, Maryland Heights, MO)		
8.Demographics				
Race/ethnicity *Infant gender+*	The biological, social and cultural traits determining individuals' social categorization.	Demographic questionnaire	Categorical (Caucasian/Black/Asian/Hispanic or Latino/ more than one/unknown/ not reported)	Self-reported by parents at enrollment
Race/ethnicity *Infant gender+*	The duration of pregnancy since initiation (last menstrual period) to birth	Medical records	Dichotomous (Male/ female)	Recorded by research staff at discharge
Birth weight *Birth HCŧ+*	Infant's first anthropometrics assessment after birth	Medical records	Continuous Raw anthropometrics transformed into the WHO growth standardized z-scores (Mean =0, SD=1)	Recorded by research staff at enrollment
Prenatal Diagnosis+	Diagnosis of CHD during the pregnancy.	Demographic questionnaire	Dichotomous Yes/no	Self-reported by parents at enrollment

Note. * Parenting Stress Index; + Bayley Scales of Infant Development-2nd Edition; + Not included in the final analyses; ** Risk Adjustment for Congenital Heart Surgery 1; ŧ Head circumference.

Table 2

Distribution of defects in the parent study's sample, N=241

Cardiac Physiology	Groups of Defects	Specific defects within group	N (%)	Cum. for group
Single Ventricle Physiology	Contruncal Defects	Dextro-Transposition of the Great Arteries (D-TGA)	1 (0.4)	4 (1.7)
		Levo-Transposition of the Great Arteries (L-TGA)	1 (0.4)	
		Double Outlet Right Ventricle (DORV)	3 (1.2)	
Single Ventricle Physiology	Contruncal Defects	Dextro-Transposition of the Great Arteries (D-TGA)	1 (0.4)	4 (1.7)
		Levo-Transposition of the Great Arteries (L-TGA)	1 (0.4)	
		Double Outlet Right Ventricle (DORV)	3 (1.2)	
	Septal Defects	Atrioventricular Canal Defect (AVCD)	1 (0.4)	1 (0.4)
	Right Sided Defects	Valvular Pulm, Atresia	5 (2.1)	15 (6.2)
		Tricuspid Atresia	10 (4.1)	
	Left Sided Defects	Hypoplastic Left Heart Syndrome (HLHS)	47 (19.5)	49 (20.3)
		Valvar Aortic Stenosis	1 (0.4)	
		Aortic Valve Atresia	1 (0.4)	
	Other Defects	Double Inlet Left Vent	8 (3.3)	8 (3.3)
Bi-Ventricle Physiology	Contruncal Defects	Tetralogy of Fallot (TOF)	15 (6.2)	59 (24.5)
		Interrupted Aortic Arch (IAA)	5 (2.1)	
		Truncus Arteriosus	2 (0.8)	
		D-TGA	34 (14.1)	
		DORV	3 (1.2)	
	Septal Defects	Aortopulmonary (AP) Window	1 (0.4)	3 (1.2)
		Ventricular Septal Defect (VSD)	2 (0.8)	
	Right Sided Defects	Valvular Pulm, Atresia	2 (0.8)	2 (0.8)
	Left Sided Defects	Valvar Aortic Stenosis	3 (1.2)	15 (6.2)
		Coarc. Of Aorta	12 (5)	
	Pulm Venous Anomalies	Total Anomalous Pulmonary Venous Return (TAPVR)	3 (1.2)	3 (1.2)
	Other Defects	Not Specified	1 (0.4)	1 (0.4)
Healthy Infants	Normal Physiology		80 (33.2)	80 (33.2)

Table 3
Demographic and clinical characteristics of the study sample, N=167.

Categorical variables	Frequency (%)
Study group	
Infants with CHD	97 (58)
Healthy infants	70 (42)
Infant gender	
Male	109 (65)
Female	58 (35)
Ethnicity	
Hispanic	12 (7)
Non-Hispanic	119 (71)
Unknown	36 (22)
Race	
White	134 (80)
Black	24 (14)
Other	5 (3)
Unknown	4 (2)
Mother's education	
High school	8 (5)
Collage	57 (34)
Post-graduate degree	33 (20)
Unreported	69 (41)
*Post-op cardiac physiology**	
Single ventricle	49 (51)
Bi-ventricle	48 (49)
*RACHS-1 score**	
Low risk (cat' 1-3)	46 (47)
High risk (cat' 4-6)	50 (52)
Missing	1 (1)
*Perinatal diagnosis**	
Yes	50 (52)
No	47 (48)
*Feeding mode at discharge**	
Oral	40 (41)
Tube assisted	36 (37)
Missing	21 (22)
*Feeding mode at 3 months**	
Oral	62 (64)
Tube assisted	10 (10)
Missing	25 (26)

Continuous variables	Mean (SD**)	Median (Range)	IQR
Birth weight, gms (n=163)	3365 (485)	3320 (2409 - 4900)	665
Gestational age, wks (n=148)	39.0 (1.5)	39 (35-42)	2
Weight at 3 mo, z-score[±] (n=116)	-.76 (1.22)	-.53 (-5.1 1.61)	1.65
Length at 3 mo, z-score (n=114)	-.42 (1.36)	-.33 (-5.45 3.53)	1.78
Head circumference at 3 mo, z-score (n=112)	-.16 (1.27)	-.10 (-3.99 2.46)	1.59
Hospital length of stay, days (n=96)	22.61 (23.61)	15 (2-159)	14.5
MDI[†] at 6 mo (n=126)	94.55 (10)	95.5 (60- 120)	13
PDI-[†] at 6 mo (n=126)	86.64 (15.43)	91 (50-129)	18
MDI at 12 months (n=137)	95.38 (10.65)	96 (66-117)	14
PDI at 12 months (n=137)	84.76 (17.09)	85 (50-117)	28

Note. *Clinical parameter measured only within the subjects group, n=97; **Standard deviation; [±] WHO growth Z-scores; [†] Bayley's Mental and Psychomotor Development Index scores.

Table 4

Descriptive statistics and comparisons for PSI subscales at 3, 6, 9, and 12 months.*

PSI subscales+	3 months visit				6 months visit				9 months visit				12 months visit			
	N	Mean (SD †)	Median	Range	N	Mean (SD)	Median	Range	N	Mean (SD)	Median	Range	N	Mean (SD)	Median	Range
Distractibility																
All infants	128	23.30 (4.38)	24	6-33	100	23.85 (4.74)	24	13-34	95	24.08 (4.88)	24	13-44	120	24.53 (4.32)	24	13-35
CHD infants	66	23.09 (4.74)	24	6-33	46	23.65 (5.01)	23	13-34	56	24.05 (4.88)	24	13-35	64	24.19 (4.75)	24	13-35
Healthy infants	62	23.53 (4.00)	24	9-33	54	24.09 (24.09)	25	15-33	39	23.62 (4.95)	23	17-31	56	24.91 (3.78)	25	17-34
Sub-sample P-value	0.571				0.647				0.942				0.362			
Adaptability																
All infants	129	24.39 (5.84)	25	1-45	100	22.80 (5.03)	23	11-36	95	23.09 (4.74)	23	12-38	120	24.10 (4.81)	24	13-38
CHD infants	66	24.97 (6.02)	25	8-45	54	22.94 (4.75)	23.5	11-34	56	23.27 (4.90)	24	12-34	64	24.25 (4.65)	24.5	13-34
Healthy infants	63	23.78 (5.63)	24	1-41	46	22.63 (5.38)	23	12-36	39	22.85 (4.56)	23	12-38	56	23.93 (5.03)	24	13-38
Sub-sample P-value	0.248				0.757				0.672				0.717			
Reinforces Parents																
All infants	128	7.91 (2.43)	17	5-22	100	7.65 (2.17)	7	6-16	95	7.97 (2.51)	7	6-16	120	7.69 (2.20)	7	6-15

CHD infants	65	8.08 (2.72)	7	5-22	54	7.57 (2.024)	7	6-14	56	7.89 (2.69)	6.5	6-16	64	7.45 (2.07)	6	6-13
Healthy infants	63	7.75 (2.09)	7	6-13	46	7.74 (2.35)	7	6-15	39	8.08 (2.25)	7	6-13	56	7.96 (2.34)	7	6-15
Sub-sample P-value	0.443				0.707				0.727				0.206			
Demandingness																
All infants	129	16.82 (5.14)	16	4-32	100	16.39 (4.49)	16	8-29	95	16.97 (4.43)	17	9-30	120	16.70 (4.45)	16	9-30
CHD infants	66	18.61 (5.65)	17.5	4-32	54	17.52 (4.58)	18	8-29	56	18.16 (4.25)	18	10-30	64	17.86 (4.46)	18.5	9-28
Healthy infants	63	14.95 (3.76)	15	7-23	46	15.07 (4.02)	15	9-26	39	15.26 (4.15)	14	9-27	56	15.38 (4.08)	16	9-30
Sub-sample P-value	<0.001				0.006				0.001				0.002			
Mood																
All infants	129	9.02 (2.90)	9	5-20	100	8.49 (2.66)	8.5	5-16	95	8.67 (2.57)	8	5-16	120	8.94 (2.50)	9	5-18
CHD infants	66	9.58 (3.04)	9	5-20	54	8.48 (2.52)	8.5	5-15	56	8.5 (2.74)	8	5-16	64	8.86 (2.35)	8.5	5-16
Healthy infants	63	8.43 (2.63)	8	5-17	46	8.5 (2.85)	8.5	5-16	39	8.92 (2.30)	10	5-14	56	9.04 (2.69)	9	5-18
Sub-sample P-value	0.024				0.973				0.432				0.702			
Acceptability																
All infants	129	10.89 (3.52)	11	2-24	100	10.90 (3.11)	11	7-19	95	11.25 (3.40)	11	7-23	120	10.98 (3.10)	11	7-18
CHD infants	66	11.32 (4.13)	11.5	2-24	54	11.50 (3.48)	11	7-19	56	11.63 (3.54)	11	7-23	64	11.03 (3.38)	11	7-18
Healthy infants	63	10.44 (2.70)	11	7-16	46	10.20 (2.46)	10	7-16	39	10.72 (3.15)	11	7-17	56	10.93 (2.78)	11	7-17
Sub-sample P-value	0.159				0.036				0.203				0.857			
Child Domain																
All infants	129	92.09 (19.39)	162	28-129	100	90.08 (16.47)	89	57-129	95	92.04 (16.32)	90	59-129	120	92.94 (15.85)	93	58-147

CHD infants	66	95.52 (21.37)	94.5	28-162	54	91.67 (16.24)	92.5	59-126	56	93.50 (16.47)	92	59-129	64	93.64 (15.67)	92.5	58-133
Healthy infants	63	88.51 (16.49)	90	38-128	46	88.22 (16.73)	88	57-129	39	89.95 (16.08)	90	60-129	56	92.14 (16.17)	93	62-147
Sub-sample P-value	0.040				0.299				0.299				0.608			
Competence																
All infants	129	23.16 (5.72)	23	7-42	100	23.64 (5.50)	23	14-38	95	23.77 (5.51)	24	13-39	120	23.23 (6.22)	22	9-49
CHD infants	66	24.17 (5.54)	24	12-42	54	23.76 (5.22)	24	16-38	56	23.66 (5.20)	24	13-36	64	23.34 (6.15)	22.5	13-41
Healthy infants	63	22.10 (5.75)	22	7-35	46	23.50 (5.88)	23	14-38	39	23.92 (5.99)	23	14-39	56	23.09 (6.36)	22	9-49
Sub-sample P-value	0.039				0.816				0.821				0.824			
Isolation																
All infants	126	12.18 (4.03)	12	5-27	100	11.95 (4.10)	12	6-27	95	12.04 (3.88)	12	6-23	119	11.82 (4.24)	12	6-27
CHD infants	66	12.55 (3.90)	13	5-27	54	12.46 (4.42)	12	6-27	56	12.20 (3.78)	12	6-20	64	12.03 (4.27)	12	6-27
Healthy infants	60	11.78 (4.16)	11	6-22	46	11.35 (3.64)	11	6-21	39	11.82 (4.05)	12	6-23	55	11.58 (4.22)	12	6-24
Sub-sample P-value	0.291				0.177				0.644				0.566			
Attachment																
All infants	126	11.13 (2.62)	11	7-21	100	10.51 (2.63)	10	7-18	95	10.52 (2.76)	10	7-22	119	10.29 (2.64)	10	7-19
CHD infants	66	11.12 (2.62)	11	8-21	54	10.48 (2.73)	10	7-18	56	10.48 (2.91)	10	7-22	64	9.86 (2.44)	9	7-18
Healthy infants	60	11.15 (2.65)	11	7-19	46	10.54 (2.54)	10	7-17	39	10.56 (2.56)	10	7-16	55	10.78 (2.80)	10	7-19
Sub-sample P-value	0.951				0.907				0.888				0.057			
Parental Health																
All infants	126	12.20 (3.06)	12	4-21	100	11.98 (3.01)	12	6-21	95	11.98 (2.84)	12	6-20	119	12.07 (3.05)	11	7-21

CHD infants	66	12.27 (2.99)	12	4-19	54	12.00 (3.02)	12	6-18	56	12.04 (2.78)	11.5	8-19	64	12.03 (3.10)	11	7-20
Healthy infants	60	12.20 (3.15)	12	6-21	46	11.96 (3.03)	11.5	7-21	39	11.90 (2.96)	12	6-20	55	12.11 (3.02)	11	7-21
Sub-sample P-value	0.776				0.943				0.817				0.890			
Role Restriction																
All infants	126	17.39 (5.01)	16.5	8-35	100	17.79 (4.64)	18	7-33	95	17.95 (4.83)	18	9-32	119	17.09 (5.16)	17	7-31
CHD infants	66	17.77 (5.09)	16	9-35	54	17.83 (4.55)	17	8-31	56	17.48 (4.91)	17.5	9-28	64	16.63 (5.53)	16	7-31
Healthy infants	60	16.97 (4.92)	17	8-27	46	17.74 (4.79)	18	7-33	39	18.62 (4.68)	18	10-32	55	17.64 (4.67)	17	9-31
Sub-sample P-value	0.369				0.920				0.262				0.288			
Depression																
All infants	125	17.34 (4.71)	17	4-30	100	16.81 (4.67)	17	9-30	95	16.92 (4.91)	17	9-33	119	16.88 (5.55)	17	8-36
CHD infants	66	17.77 (4.16)	17	9-28	54	16.63 (4.46)	17	9-30	56	16.73 (4.43)	16.5	9-29	64	17.09 (5.86)	17	8-36
Healthy infants	59	17.08 (5.25)	16	8-30	46	17.02 (4.95)	17.5	9-30	39	17.00 (5.15)	18	9-29	55	16.64 (5.20)	17	9-36
Sub-sample P-value	0.283				0.678				0.787				0.656			
Spouse																
All infants	125	16.22 (4.80)	16	7-30	100	16.70 (5.04)	16	7-33	95	16.41 (5.29)	16	7-32	119	15.61 (4.68)	15	7-32
CHD infants	66	16.77 (5.04)	17	7-30	54	16.70 (5.11)	15	7-33	56	16.64 (5.62)	16	7-32	64	15.56 (5.014)	15	7-32
Healthy infants	59	15.63 (4.50)	16	7-25	46	16.70 (5.00)	16.5	8-30	39	16.08 (4.81)	15	7-26	55	15.65 (4.30)	16	7-25
Sub-sample P-value	0.178				0.994				0.610				0.915			
Parent Domain																
All infants	126	109.95 (21.22)	109.5	66-178	100	109.38 (21.86)	109.5	66-177	95	109.58 (22.14)	112	66-179	119	107.10 (24.06)	105	60-196

CHD infants	66	112.42 (20.76)	109.5	66-178	54	109.87 (20.59)	109	68-157	56	109.23 (20.69)	112.5	68-151	64	106.55 (25.61)	105	60-196
Healthy infants	60	107.23 (21.57)	108.5	67-161	46	108.80 (23.48)	109.5	66-177	39	110.08 (24.33)	110	66-179	55	107.75 (22.34)	105	72-196
Sub-sample P-value	0.171				0.809				0.856				0.788			
Total Stress																
All infants	126	202.18 (36.61)	201	129-340	100	199.46 (34.29)	200.5	134-305	95	201.62 (35.69)	203	131-308	119	200.15 (36.69)	197	136-343
CHD infants	66	207.94 (38.04)	205	133-340	54	201.54 (32.13)	204.5	136-267	56	202.73 (34.44)	207.5	131-275	64	200.19 (37.35)	195.5	136-319
Healthy infants	60	195.85 (34.17)	194.5	129-280	46	197.02 (36.88)	197.5	134-305	39	200.03 (37.81)	199	148-308	55	200.11 (36.24)	203	147-343
Sub-sample P-value	0.064				0.514				0.718				0.991			
Life Stress																
All infants	127	10.16 (8.25)	8	0-42	100	8.75 (8.40)	7	0-40	95	8.99 (8.50)	7	0-34	119	8.76 (7.43)	8	0-39
CHD infants	66	9.65 (7.41)	8	0-35	54	8.56 (7.85)	8	0-28	56	10.32 (8.32)	9	0-33	64	10.13 (8.03)	8.5	0-40
Healthy infants	61	10.70 (9.11)	9	0-42	46	8.98 (9.10)	6	0-40	39	7.08 (8.51)	4	0-34	55	7.2 (6.46)	7	0-19
Sub-sample P-value	0.475				0.804				0.067				0.033			

Note. *Two sample t-test for comparing PSI subscale by group (CHD vs. healthy); [+]Parenting Stress Index subscales; [†] Standard Deviation.

Table 5a

Final Mixed-Effects model results[†] for PSI subscales regressed on Visit, CHD/healthy infant, and Visit x CHD/healthy infant terms.

PSI subscale[*]	Parameter	β	SE[**]	95% CI[‡]	P	N
Distractibility						
	Visit[+]	0.43	0.25	(-0.06, 0.92)	0.086	124
	CHD infants[±]	-0.44	1.42	(-3.22, 2.35)	0.758	
	Visit x CHD/healthy	0.05	0.35	(-0.64, 0.74)	0.892	
Adaptability						
	Visit	0.19	0.25	(-0.30, 0.68)	0.449	124
	CHD infants	2.00	1.55	(-1.02, 5.03)	0.195	
	Visit x CHD/healthy	-0.34	0.35	(-1.02, 0.35)	0.337	
Reinforces parents						
	Visit	0.17	0.11	(-0.06, 0.39)	0.144	124
	CHD infants	0.92	0.65	(-0.36, 2.20)	0.16	
	Visit x CHD/healthy	-0.32	0.16	(-0.64, -0.01)	0.044	
Demandingness						
	Visit	0.16	0.24	(-0.30, 0.63)	0.491	124
	CHD infants	4.44	1.39	(1.71, 7.17)	0.001	
	Visit x CHD/healthy	-0.38	0.33	(-1.03, 0.27)	0.246	
Mood						
	Visit	0.24	0.14	(-0.03, 0.50)	0.083	124
	CHD infants	2.00	0.81	(0.41, 3.59)	0.014	
	Visit x CHD/healthy	-0.49	0.19	(-0.87, -0.12)	0.010	

Acceptability						
	Visit	0.11	0.17	(-0.23, 0.45)	0.53	124
	CHD infants	1.11	0.97	(-0.79, 3.01)	0.253	
	Visit x CHD/healthy	-0.14	0.24	(-0.62, 0.34)	0.568	
Child domain						
	Visit	1.33	0.81	(-0.25, 2.91)	0.099	124
	CHD infants	9.64	5.05	(-0.25, 19.53)	0.056	
	Visit x CHD/healthy	-1.55	1.13	(-3.77, 0.67)	0.171	
Competence						
	Visit	0.49	0.24	(0.01, 0.96)	0.043	124
	CHD infants	3.56	1.47	(0.67, 6.44)	0.016	
	Visit x CHD/healthy	-0.67	0.34	(-1.34, 0.00)	0.051	
Isolation						
	Visit	0.06	0.16	(-0.25, 0.37)	0.719	123
	CHD infants	2.04	0.98	(0.12, 3.96)	0.038	
	Visit x CHD/healthy	-0.21	0.22	(-0.65, 0.22)	0.336	
Attachment						
	Visit	-0.08	0.11	(-0.31, 0.14)	0.456	123
	CHD infants	1.012	0.71	(-0.38, 2.40)	0.154	
	Visit x CHD/healthy	-0.39	0.16	(-0.70, -0.08)	0.015	
Health						
	Visit	-0.02	0.15	(-0.31, 0.27)	0.874	123
	CHD infants	1.19	0.91	(-0.60, 2.98)	0.191	
	Visit x CHD/healthy	-0.18	0.21	(-0.59, 0.22)	0.380	
Role restriction						
	Visit	0.30	0.21	(-0.12, 0.71)	0.162	123

	CHD infants	2.67	1.29	(0.15, 5.19)	0.038	
	Visit x CHD/healthy	-0.70	0.30	(-1.28, -0.11)	0.019	
Depression						
	Visit	0.02	0.21	(-0.38, 0.43)	0.911	123
	CHD infants	1.95	1.23	(-0.46, 4.37)	0.113	
	Visit x CHD/healthy	-0.34	0.29	(-0.90, 0.23)	0.245	
Spouse						
	Visit	-0.12	0.20	(-0.50, 0.26)	0.535	123
	CHD infants	0.28	1.23	(-2.13, 2.69)	0.819	
	Visit x CHD/healthy	-0.06	0.27	(-0.60, 0.48)	0.825	
Parent Domain						
	Visit	0.46	0.76	(-1.04, 1.95)	0.551	123
	CHD infants	12.13	4.88	(2.57, 21.69)	0.013	
	Visit x CHD/healthy	-2.41	1.08	(-4.53, -0.29)	0.026	
Total stress						
	Visit	1.70	1.27	(-0.79, 4.18)	0.181	123
	CHD infants	20.52	8.31	(4.22, 36.82)	0.014	
	Visit x CHD/healthy	-3.69	1.79	(-7.20, -0.18)	0.039	
Life stress						
	Visit	-1.40	0.37	(-2.12, -0.67)	0.000	123
	CHD infants	-4.38	2.47	(-9.23, 0.47)	0.076	
	Visit x CHD/healthy	1.48	0.52	(0.47, 2.49)	0.004	

Note. [†]All models are adjusted for infant length Z-scores and birthweight; [+]Visits represent the independent variable of time (continuous); [±]CHD vs. Healthy infants; [*]Parenting Stress Index subscales scores as the outcome of interest, each represents a separate multivariable model; ** Standard Error; [Ŧ]95% Confidence intervals.

Table 5b

Mixed Effects model results[†] for PSI subscales regressed on Visit[+].

	Healthy infants					CHD infants				
PSI subscale[*]	**β[+]**	**SE[**]**	**95% CI[‡]**	**P**	**N**	**β[+]**	**SE[**]**	**95% CI[‡]**	**P**	**N**
Distractibility	0.45	0.20	(0.06, 0.84)	0.023	53	0.49	0.28	(-0.06, 1.05)	0.083	71
Adaptability	0.21	0.21	(-0.20, 0.62)	0.310	53	-0.06	0.27	(-0.59, 0.47)	0.831	71
Reinforces parents	0.16	0.11	(-0.05, 0.37)	0.128	53	-0.18	0.12	(-0.42, 0.05)	0.128	71
Demandingness	0.18	0.20	(-0.21, 0.57)	0.363	53	-0.12	0.26	(-0.63, 0.39)	0.644	71
Mood	0.23	0.12	(-0.01, 0.47)	0.060	53	-0.25	0.15	(-0.54, 0.03)	0.082	71
Acceptability	0.12	0.14	(-0.15, 0.39)	0.389	53	0.02	0.20	(-0.37, 0.40)	0.938	71
Child domain	1.36	0.64	(0.11, 2.62)	0.033	53	-0.04	0.91	(-1.82, 1.74)	0.965	71
Competence	0.48	0.26	(-0.03, 0.99)	0.065	53	-0.20	0.22	(-0.64, 0.24)	0.375	71
Isolation	0.06	0.15	(-0.23, 0.35)	0.698	52	-0.14	0.17	(-0.47, 0.19)	0.404	71
Attachment	-0.09	0.11	(-0.31, 0.12)	0.386	52	-0.49	0.12	(-0.73, -0.25)	0.000	71
Health	-0.02	0.13	(-0.29, 0.24)	0.864	52	-0.17	0.16	(-0.48, 0.13)	0.270	71
Role restriction	0.30	0.22	(-0.14, 0.74)	0.179	52	-0.38	0.20	(-0.78, 0.02)	0.059	71
Depression	0.01	0.23	(-0.45, 0.46)	0.977	52	-0.27	0.23	(-0.72, 0.19)	0.250	71
Spouse	-0.11	0.16	(-0.43, 0.21)	0.494	52	-0.18	0.22	(-0.62, 0.25)	0.415	71
Parent domain	0.46	0.67	(-0.86, 1.77)	0.496	52	-1.90	0.85	(-3.57, -0.24)	0.025	71
Total stress	1.741	1.09	(-0.40, 3.88)	0.111	52	-1.84	1.41	(-4.61, 0.92)	0.192	71
Life stress	-1.38	0.42	(-2.20, -0.55)	0.001	52	0.19	0.31	(-0.42, 0.80)	0.532	71

Note. [†]All models are adjusted for infant length Z-scores and birthweight; [+] Estimates in table correspond to main effect of "Visit"; Visit represents the independent variable of time (continuous); [*] Parenting Stress Index subscales scores as the outcome of interest, each represents a separate multivariable model within each group, CHD and healthy; [**] Standard Error; [‡]95% Confidence intervals.

Table 5c

Final Mixed-Effects model results[†] for PSI subscales regressed on Visit[+], Post-op Cardiac Physiology, and Visit x Post-op Cardiac Physiology terms.

PSI subscale[*]		**β**	**SE[**]**	**95% CI[‡]**	**P**	**N**
Distractibility						
	Visit	-0.38	0.38	(-1.13, 0.37)	0.320	69
	BV physiology[±]	-6.37	2.05	(-10.39, -2.35)	0.002	
	Visit x Group	1.63	0.51	(0.62, 2.63)	0.002	
Adaptability						
	Visit	-0.57	0.39	(-1.34, 0.20)	0.145	69
	BV infants	-2.85	2.37	(-7.49, 1.80)	0.230	
	Visit x Group	0.88	0.52	(-0.14, 1.90)	0.090	
Reinforces Parents						
	Visit	-0.27	0.18	(-0.62, 0.08)	0.126	69
	BV infants	-0.16	1.06	(-2.23, 1.91)	0.878	
	Visit x Group	0.06	0.23	(-0.40, 0.51)	0.806	
Demandingness						
	Visit	-0.46	0.38	(-1.21, 0.29)	0.227	69
	BV infants	-1.74	2.27	(-6.19, 2.71)	0.443	
	Visit x Group	0.54	0.51	(-0.46, 1.55)	0.290	
Mood						
	Visit	-0.64	0.20	(-1.04, -0.24)	0.002	69
	BV infants	-2.08	1.23	(-4.50, 0.34)	0.092	
	Visit x Group	0.71	0.27	(0.17, 1.24)	0.009	

Acceptability						
	Visit	-0.22	0.29	(-0.79, 0.35)	0.450	69
	BV infants	-1.63	1.62	(-4.80, 1.54)	0.313	
	Visit x Group	0.26	0.39	(-0.51, 1.03)	0.511	
Child domain						
	Visit	-2.34	1.30	(-4.89, 0.20)	0.071	69
	BV infants	-14.16	8.15	(-30.14, 1.81)	0.082	
	Visit x Group	3.94	1.73	(0.54, 7.34)	0.023	
Competence						
	Visit	-0.54	0.33	(-1.17, 0.10)	0.100	69
	BV infants	-2.21	1.90	(-5.93, 1.52)	0.245	
	Visit x Group	0.41	0.43	(-0.44, 1.25)	0.346	
Isolation						
	Visit	-0.16	0.24	(-0.64, 0.32)	0.507	69
	BV infants	-0.19	1.39	(-2.90, 2.53)	0.893	
	Visit x Group	-0.08	0.32	(-0.72, 0.55)	0.794	
Attachment						
	Visit	-0.60	0.18	(-0.95, -0.24)	0.001	69
	BV infants	-0.67	1.07	(-2.78, 1.43)	0.532	
	Visit x Group	0.13	0.24	(-0.34, 0.59)	0.589	
Health						
	Visit	-0.18	0.23	(-0.64, 0.27)	0.425	69
	BV infants	0.36	1.34	(-2.27, 2.99)	0.788	
	Visit x Group	-0.06	0.31	(-0.66, 0.54)	0.842	
Role restriction						
	Visit	-0.57	0.30	(-1.15, 0.01)	0.054	69

	BV infants	-0.37	1.72	(-3.74, 2.99)	0.828	
	Visit x Group	0.22	0.39	(-0.54, 0.98)	0.571	
Depression						
	Visit	-0.47	0.33	(-1.12, 0.19)	0.162	69
	BV infants	-0.90	1.69	(-4.21, 2.42)	0.596	
	Visit x Group	0.25	0.44	(-0.62, 1.12)	0.569	
Spouse						
	Visit	-0.31	0.32	(-0.94, 0.32)	0.34	69
	BV infants	-0.93	1.74	(-4.35, 2.49)	0.593	
	Visit x Group	0.06	0.43	(-0.78, 0.90)	0.885	
Parent domain						
	Visit	-2.78	1.23	(-5.18, -0.38)	0.023	69
	BV infants	-5.09	6.63	(-18.09, 7.90)	0.442	
	Visit x Group	0.84	1.62	(-2.34, 4.02)	0.604	
Total stress						
	Visit	-5.07	2.01	(-9.01, -1.12)	0.012	69
	BV infants	-19.75	12.31	(-43.88, 4.37)	0.109	
	Visit x Group	4.80	2.65	(-0.40, 10.00)	0.071	
Life stress						
	Visit	-0.38	0.45	(-1.25, 0.49)	0.392	69
	BV infants	-6.49	2.68	(-11.75, -1.24)	0.015	
	Visit x Group	0.93	0.58	(-0.21, 2.08)	0.109	

Note. [†]All models are adjusted for infant length Z-scores, birthweight, and feeding mode at discharge (exclusively oral feeding vs. device assisted feeding); [+]Visits represent the independent variable of time (continuous); [±]Bi-ventricle vs. Single-ventricle post-op cardiac physiology; [*]Parenting Stress Index subscales scores as the outcome of interest, each represents a separate multivariable model; [**] Standard Error; [‡]95% Confidence intervals

Table 5d

Mixed Effects model results[†] for PSI subscales regressed on Visit[+].

	Single Ventricle Physiology					Bi-Ventricle Physiology				
PSI subscale[*]	**β[+]**	**SE[**]**	**95% CI[‡]**	**P**	**N**	**β[+]**	**SE[**]**	**95% CI[‡]**	**P**	**N**
Distractibility	-0.26	0.39	(-0.98, 0.46)	0.476	34	1.13	0.38	(0.37, 1.88)	0.003	35
Adaptability	-0.56	0.45	(-1.43, 0.33)	0.221	34	0.28	0.33	(-0.37, 0.93)	0.403	35
Reinforces parents	-0.17	0.21	(-0.57, 0.24)	0.412	34	-0.28	0.15	(-0.57, 0.01)	0.061	35
Demandingness	-0.45	0.44	(-1.31, 0.42)	0.310	34	0.01	0.32	(-0.61, 0.63)	0.978	35
Mood	-0.59	0.26	(-1.11, -0.07)	0.026	34	0.02	0.14	(-0.27, 0.30)	0.908	35
Acceptability	-0.16	0.35	(-0.86, 0.53)	0.643	34	-0.03	0.23	(-0.48, 0.41)	0.885	35
Child domain	-2.00	1.52	(-4.98, 0.98)	0.188	34	1.17	1.05	(-0.89, 3.24)	0.265	35
Competence	-0.54	0.41	(-1.35, 0.26)	0.186	34	-0.23	0.30	(-0.81, 0.36)	0.451	35
Isolation	-0.10	0.26	(-0.61, 0.40)	0.684	34	-0.33	0.24	(-0.80, 0.13)	0.161	35
Attachment	-0.61	0.21	(-1.02, -0.19	0.004	34	-0.47	0.15	(-0.77, -0.17)	0.002	35
Health	-0.08	0.22	(-0.51, 0.35)	0.711	34	-0.31	0.24	(-0.79, 0.17)	0.208	35
Role restriction	-0.61	0.31	(-1.21, -0.02)	0.043	34	-0.41	0.31	(-1.02, 0.19)	0.181	35
Depression	-0.36	0.36	(-1.09, 0.34)	0.303	34	-0.34	0.30	(-0.92, 0.25)	0.258	35
Spouse	-0.35	0.32	(-0.99, 0.28)	0.274	34	-0.31	0.31	(-0.91, 0.30)	0.321	35
Parent domain	-2.53	1.25	(-4.98, -0.08)	0.043	34	-2.23	1.29	(-4.76, 0.30)	0.083	35
Total stress	-4.51	2.09	(-8.61, -0.41)	0.031	34	-1.07	1.86	(-4.72, 2.59)	0.567	35
Life stress	-0.40	0.50	(-1.38, 0.58)	0.421	34	0.47	0.40	(-0.31, 1.25)	0.240	35

Note. [†]All models are adjusted for infant length Z-scores, birthweight, and feeding mode at discharge (exclusively oral feeding vs. device assisted feeding); [+] Estimates in table correspond to main effect of "Visit"; [+]Visit represents the independent variable of time (continuous); [*] Parenting Stress Index subscales scores as the outcome of interest, each represents a separate multivariate model within each group, Single-ventricle and Bi-ventricle physiology; [**] Standard Error; [‡]95% Confidence intervals.

Table 6a

Multivariable[†] regression models for MDI[+] at 6 months on PSI at 3 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI**	**P**	**R^2**
Distractibility	39	0.38	0.26	(-0.15, 0.92)	0.153	0.32	34	0.18	0.29	(-0.40, 0.77)	0.528	0.40
Adaptability	39	-0.01	0.25	(-0.51, 0.49)	0.977	0.28	34	0.05	0.23	(-0.42, 0.53)	0.821	0.40
Reinforces parents	39	0.09	0.58	(-1.09, 1.27)	0.880	0.28	34	0.14	0.49	(-0.85, 1.14)	0.769	0.40
Demandingness	39	-0.21	0.33	(-0.87, 0.46)	0.536	0.29	34	0.11	0.24	(-0.39, 0.61)	0.659	0.40
Mood	39	0.19	0.48	(-0.79, 1.17)	0.696	0.28	34	0.18	0.57	(-0.99, 1.34)	0.760	0.40
Acceptability	39	0.20	0.47	(-0.74, 1.14)	0.669	0.28	34	0.54	0.47	(-0.42, 1.49)	0.257	0.42
Child domain	39	0.03	0.08	(-0.13, 0.19)	0.688	0.28	34	0.04	0.07	(-0.09, 0.18)	0.530	0.40
Competence	39	0.09	0.22	(-0.35, 0.53)	0.682	0.28	34	-0.20	0.24	(-0.69, 0.28)	0.396	0.41
Isolation	37	0.18	0.31	(-0.44, 0.81)	0.554	0.28	34	-0.42	0.46	(-1.36, 0.52)	0.366	0.41
Attachment	37	-0.18	0.49	(-1.19, 0.83)	0.717	0.27	34	-0.20	0.54	(-1.32, 0.91)	0.712	0.40
Health	37	0.05	0.44	(-0.84, 0.94)	0.913	0.27	34	-1.00	0.45	(-1.92, -0.08)	0.034	0.48
Role restriction	37	0.07	0.26	(-0.46, 0.60)	0.782	0.27	34	-0.11	0.32	(-0.77, 0.55)	0.730	0.40
Depression	37	0.01	0.26	(-0.51, 0.54)	0.958	0.27	34	-0.63	0.43	(-1.51, 0.25)	0.152	0.44
Spouse	37	0.50	0.24	(0.00, 0.99)	0.048	0.35	34	0.06	0.44	(-0.85, 0.97)	0.892	0.40
Parent domain	37	0.05	0.06	(-0.07, 0.16)	0.439	0.28	34	-0.10	0.08	(-0.27, 0.07)	0.218	0.43
Total stress	37	0.03	0.04	(-0.05, 0.10)	0.485	0.28	34	-0.01	0.04	(-0.10, 0.08)	0.824	0.40
Life stress	38	0.03	0.12	(-0.22, 0.28)	0.803	0.28	34	0.01	0.21	(-0.41, 0.43)	0.952	0.40

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Mental Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6b

Multivariable[†] regression models for MDI[+] at 6 months on PSI at 6 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	33	-0.26	0.32	(-0.92, 0.41)	0.437	0.31	41	-0.03	0.33	(-0. 70, 0.64)	0.936	0.31
Adaptability	33	-0.19	0.24	(-0.67, 0.29)	0.428	0.31	41	-0.54	0.38	(-1.32, 0.23)	0.165	0.35
Reinforces parents	33	-0.93	0.56	(-2.09, 0.20)	0.103	0.36	41	-1.57	0.76	(-3.11, -0.02)	0.047	0.38
Demandingness	33	-0.30	0.29	(-0.89, 0.29)	0.307	0.32	41	0.06	0.33	(-0.61, 0.73)	0.855	0.31
Mood	33	-0.21	0.48	(-1.20, 0.77)	0.658	0.30	41	0.28	0.62	(-0.97, 1.53)	0.650	0.31
Acceptability	33	0.25	0.51	(-0.78, 1.28)	0.624	0.30	41	0.19	0.43	(-0.69, 1.06)	0.667	0.31
Child domain	33	-0.07	0.08	(-0.23, 0.08)	0.342	0.31	41	-0.04	0.10	(-0.25, 0.17)	0.692	0.31
Competence	33	0.14	0.22	(-0.30, 0.59)	0.517	0.30	41	-0.45	0.25	(-0.95, 0.05)	0.076	0.37
Isolation	33	-0.44	0.33	(-1.12, 0.24)	0.196	0.33	41	-0.71	0.35	(-1.42, -0.01)	0.047	0.38
Attachment	33	-0.22	0.47	(-1.19, 0.74)	0.639	0.30	41	-0.58	0.49	(-1.57, 0.41)	0.242	0.34
Health	33	0.19	0.40	(-0.63, 1.01)	0.634	0.30	41	-0.64	0.45	(-1.55, 0.27)	0.161	0.35
Role restriction	33	0.12	0.24	(-0.37, 0.61)	0.630	0.30	41	-0.31	0.34	(-1.00, 0.38)	0.369	0.33
Depression	33	-0.29	0.23	(-0.75, 0.18)	0.223	0.33	41	-0.67	0.29	(-1.26, -0.08)	0.028	0.40
Spouse	33	0.17	0.23	(-0.29, 0.64)	0.454	0.31	41	-0.00	0.32	(-0.65, 0.64)	0.996	0.31
Parent domain	33	-0.00	0.05	(-0.10, 0.10)	0.981	0.30	41	-0.14	0.07	(-0.28, -0.00)	0.044	0.40
Total stress	33	-0.01	0.03	(-0.08, 0.05)	0.683	0.30	41	-0.07	0.05	(-0.16, 0.02)	0.136	0.35
Life stress	33	-0.03	0.13	(-0.29, 0.23)	0.822	0.29	41	0.02	0.19	(-0.35, 0.40)	0.903	0.31

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Mental Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6c:

Multivariable[†] regression models for PDI[+] at 6 months on PSI at 3 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	38	0.87	0.40	(0.06, 1.67)	0.036	0.14	35	0.64	0.49	(-0.37, 1.64)	0.206	0.25
Adaptability	38	0.36	0.38	(-0.41, 1.13)	0.347	0.05	35	0.67	0.39	(-0.14, 1.47)	0.102	0.28
Reinforces parents	38	-0.54	0.91	(-2.40, 1.31)	0.555	0.03	34	1.60	0.76	(0.04, 3.17)	0.045	0.32
Demandingness	38	-0.45	0.52	(-1.50, 0.61)	0.399	0.03	35	0.52	0.42	(-0.33, 1.38)	0.219	0.25
Mood	38	-0.85	0.74	(-2.35, 0.65)	0.257	0.06	35	1.55	0.90	(-0.29, 3.39)	0.095	0.28
Acceptability	38	-0.13	0.75	(-1.66, 1.41)	0.869	0.02	35	1.28	0.80	(-0.35, 2.91)	0.119	0.27
Child domain	38	0.05	0.12	(-0.20, 0.29)	0.691	0.03	35	0.22	0.11	(-0.01, 0.45)	0.055	0.30
Competence	38	0.33	0.34	(-0.36, 1.01)	0.340	0.05	35	-0.15	0.42	(-1.01, 0.71)	0.724	0.21
Isolation	36	0.76	0.46	(-0.18, 1.70)	0.108	0.10	35	-0.45	0.82	(-2.11, 1.22)	0.588	0.22
Attachment	36	-0.41	0.78	(-2.00, 1.17)	0.600	0.03	35	0.28	0.95	(-1.66, 2.21)	0.773	0.21
Health	36	0.36	0.691	(-1.05,1.77)	0.609	0.03	35	-0.32	0.86	(-2.07, 1.43)	0.711	0.21
Role restriction	36	0.54	0.42	(-0.33, 1.40)	0.214	0.07	35	-0.74	0.54	(-1.84, 0.36)	0.178	0.26
Depression	36	0.28	0.40	(-0.54, 1.09)	0.490	0.04	35	0.55	0.74	(-0.96, 2.07)	0.463	0.22
Spouse	36	0.62	0.39	(-0.18, 1.41)	0.123	0.10	35	-0.57	0.78	(-2.16, 1.02)	0.471	0.22
Parent domain	36	0.12	0.09	(-0.06, 0.31)	0.177	0.08	35	-0.09	0.15	(-0.40, 0.22)	0.551	0.22
Total stress	36	0.06	0.06	(-0.05, 0.18)	0.287	0.06	35	0.07	0.08	(-0.08, 0.23)	0.351	0.23
Life stress	37	0.13	0.19	(-0.26, 0.52)	0.503	0.04	35	-0.50	0.35	(-1.22, 0.21)	0.162	0.23

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Psychomotor Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6d

Multivariable[†] regression models for PDI[+] at 6 months on PSI at 6 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	32	0.35	0.60	(-0.89, 1.58)	0.570	0.05	42	0.58	0.48	(-0.39, 1.55)	0.231	0.23
Adaptability	32	-0.11	0.44	(-1.01, 0.79)	0.808	0.04	42	-0.01	0.58	(-1.19, 1.17)	0.989	0.20
Reinforces parents	32	-2.21	1.00	(-4.25, -0.16)	0.035	0.19	42	0.52	1.17	(-1.84, 2.88)	0.658	0.20
Demandingness	32	-0.28	0.54	(-1.38, 0.82)	0.603	0.05	42	-0.02	0.48	(-1.00, 0.95)	0.962	0.20
Mood	32	-1.21	0.87	(-2.98, 0.57)	0.175	0.10	42	1.53	0.88	(-0.26, 3.31)	0.091	0.26
Acceptability	32	-0.27	0.93	(-2.18, 1.65)	0.778	0.05	42	0.04	0.62	(-1.22, 1.30)	0.953	0.20
Child domain	32	-0.09	0.14	(-0.39, 0.20)	0.523	0.07	42	0.11	0.15	(-2.00, 0.41)	0.483	0.21
Competence	32	0.16	0.40	(-0.67, 0.98)	0.704	0.05	42	-0.26	0.37	(-1.02, 0.49)	0.481	0.21
Isolation	32	-0.43	0.63	(-1.73, 0.86)	0.500	0.06	42	-0.70	0.53	(-1.77, 0.38)	0.197	0.24
Attachment	32	-1.23	0.86	(-3.00, 0.54)	0.165	0.11	42	1.36	0.70	(-0.06, 2.78)	0.059	0.27
Health	32	0.41	0.74	(-1.10, 1.92)	0.579	0.05	42	-0.09	0.67	(-1.45, 1.27)	0.894	0.20
Role restriction	32	-0.02	0.44	(-0.93, 0.88)	0.959	0.04	42	-0.74	0.49	(-1.73, 0.24)	0.135	0.25
Depression	32	-0.20	0.43	(-1.08, 0.69)	0.654	0.05	42	0.10	0.46	(-0.83, 1.04)	0.824	0.20
Spouse	32	0.04	0.42	(-0.82, 0.91)	0.924	0.04	42	-0.20	0.47	(-1.15, 0.75)	0.676	0.20
Parent domain	32	-0.02	0.09	(-0.20, 0.17)	0.868	0.04	42	-0.06	0.11	(-0.28, 0.16)	0.568	0.21
Total stress	32	-0.02	0.06	(-0.15, 0.10)	0.708	0.05	42	-0.00	0.07	(-0.15, 0.14)	0.961	0.20
Life stress	32	-0.16	0.23	(-0.63, 0.32)	0.511	0.06	42	-0.33	0.27	(-0.87, 0.21)	0.218	0.23

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Psychomotor Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6e

Multivariable[†] regression models for MDI[+] at 12 months on PSI at 3 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	39	0.38	0.31	(-0.24, (1.01)	0.221	0.12	50	-0.18	0.35	(-0.88, 0.53)	0.616	0.23
Adaptability	39	-0.10	0.28	(-0.68, 0.48)	0.723	0.09	50	0.02	0.28	(-0.55, 0.60)	0.940	0.23
Reinforces parents	39	-0.65	0.66	(-1.99, 0.70)	0.334	0.11	49	-0.25	0.62	(-1.50, 1.00)	0.688	0.24
Demandingness	39	-0.11	0.38	(-0.87, 0.66)	0.772	0.09	50	-0.45	0.31	(-1.07, 0.18)	0.159	0.30
Mood	39	-0.55	0.55	(-1.67, 0.57)	0.324	0.11	50	-0.72	0.56	(-1.84, 0.40)	0.202	0.25
Acceptability	39	0.42	0.54	(-0.66, 1.51)	0.433	0.10	50	-0.35	0.46	(-1.29, 0.59)	0.457	0.24
Child domain	39	0.00	0.09	(-0.18, 0.18)	0.992	0.09	50	-0.06	0.08	(-0.23, 0.10)	0.445	0.24
Competence	39	0.26	0.25	(-0.24, 0.77)	0.298	0.11	50	-0.35	0.31	(-0.96, 0.27)	0.264	0.25
Isolation	37	0.23	0.35	(-0.47, 0.94)	0.505	0.10	50	0.08	0.49	(-0.98, 1.00)	0.987	0.23
Attachment	37	-0.28	0.56	(-1.42, 0.87)	0.628	0.09	50	-0.76	0.60	(-1.97, 0.44)	0.209	0.25
Health	37	0.23	0.50	(-0.78, 1.25)	0.640	0.09	50	0.09	0.54	(-1.00, 1.19)	0.863	0.23
Role restriction	37	-0.35	0.29	(-0.94, 0.24)	0.236	0.12	50	-0.05	0.35	(-0.75, 0.65)	0.878	0.23
Depression	37	-0.16	0.29	(-0.75, 0.44)	0.596	0.09	50	-0.56	0.41	(-1.38, 0.26)	0.178	0.26
Spouse	37	-0.03	0.29	(-0.63, 0.57)	0.911	0.08	50	-0.57	0.45	(-1.48, 0.34)	0.217	0.25
Parent domain	37	-0.00	0.07	(-0.14, 0.14)	0.983	0.08	50	-0.10	0.09	(-0.28, 0.08)	0.281	0.25
Total stress	37	-0.00	0.04	(-0.09, 0.08)	0.929	0.08	50	-0.05	0.05	(-0.15, 0.05)	0.295	0.24
Life stress	38	-0.05	0.14	(-0.34, 0.24)	0.727	0.09	50	-0.08	0.22	(-0.51, 0.36)	0.729	0.23

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Mental Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6f

Multivariable[†] regression models for MDI[+] at 12 months on PSI at 6 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	33	-0.65	0.40	(-1.45, 0.16)	0.113	0.25	44	0.23	0.42	(-0.62, 1.07)	0.591	0.27
Adaptability	33	-0.59	0.28	(-1.16, -0.02)	0.044	0.29	44	-0.11	0.49	(-1.11, 0.89)	0.826	0.26
Reinforces parents	33	-2.10	0.62	(-3.37, -0.84)	0.002	0.41	44	-0.33	1.01	(-2.37, 1.71)	0.745	0.26
Demandingness	33	-0.55	0.36	(-1.28, 0.18)	0.135	0.24	44	-0.21	0.39	(-1.00, 0.59)	0.601	0.27
Mood	33	-0.51	0.60	(-1.73, 0.72)	0.402	0.20	44	-0.09	0.77	(-1.64, 1.46)	0.907	0.26
Acceptability	33	-0.59	0.63	(-1.88, 0.70)	0.356	0.20	44	-0.44	0.53	(-1.52, 0.64)	0.416	0.27
Child domain	33	-0.21	0.09	(-0.39, -0.02)	0.030	0.30	44	-0.04	0.13	(-0.30, 0.22)	0.749	0.26
Competence	33	0.15	0.28	(-0.42, 0.71)	0.601	0.18	44	-0.20	0.32	(-0.84, 0.44)	0.531	0.27
Isolation	33	-0.15	0.43	(-1.03, 0.73)	0.733	0.18	44	-0.35	0.47	(-1.29, 0.59)	0.455	0.27
Attachment	33	-1.15	0.56	(-2.29, -0.01)	0.048	0.28	44	-0.25	0.63	(-1.52, 1.03)	0.696	0.26
Health	33	0.11	0.51	(-0.93, 1.14)	0.830	0.18	44	-0.15	0.58	(-1.32, 1.01)	0.793	0.26
Role restriction	33	-0.25	0.30	(-0.86, 0.36)	0.414	0.20	44	-0.41	0.42	(-1.27, 0.45)	0.337	0.28
Depression	33	-0.21	0.29	(-0.80, 0.39)	0.489	0.19	44	-0.13	0.40	(-0.94, 0.68)	0.746	0.26
Spouse	33	-0.29	0.29	(-0.88, 0.29)	0.312	0.21	44	0.02	0.41	(-0.80, 0.85)	0.954	0.26
Parent domain	33	-0.04	0.06	(-0.17, 0.09)	0.523	0.19	44	-0.07	0.09	(-0.25, 0.12)	0.480	0.27
Total stress	33	-0.05	0.04	(-0.13, 0.03)	0.192	0.22	44	-0.03	0.06	(-0.16, 0.09)	0.539	0.27
Life stress	33	-0.15	0.16	(-0.48, 0.17)	0.339	0.20	44	-0.36	0.23	(-0.82, 0.10)	0.121	0.31

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Mental Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6g

Multivariable[†] regression models for MDI[+] at 12 months on PSI at 9 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	27	0.18	0.49	(-0.84, 1.19)	0.725	0.11	30	0.54	0.39	(-0.26. 1.34)	0.175	0.48
Adaptability	27	0.04	0.37	(-0.73, 0.81)	0.907	0.10	30	0.34	0.36	(-0.40. 1.08)	0.348	0.46
Reinforces parents	27	0.99	0.84	(-0.75, 2.72)	0.251	0.15	30	-0.69	0.69	(-2.11. 0.72)	0.323	0.46
Demandingness	27	0.57	0.40	(-0.25, 1.39)	0.166	0.18	30	0.04	0.40	(-0.78. 0.86)	0.920	0.44
Mood	27	0.90	0.78	(-0.72, 2.52)	0.261	0.15	30	-0.55	0.69	(-1.97, 0.87)	0.433	0.46
Acceptability	27	0.90	0.58	(-0.29, 2.10)	0.130	0.19	30	-0.07	0.53	(-1.17, 1.03)	0.897	0.44
Child domain	27	0.12	0.11	(-0.10, 0.34)	0.263	0.15	30	0.05	0.12	(-0.19, 0.29)	0.692	0.45
Competence	27	0.45	0.26	(-0.08, 0.99)	0.092	0.21	30	-0.01	0.35	(-0.73, 0.72)	0.988	0.44
Isolation	27	0.55	0.40	(-0.29, 1.38)	0.190	0.17	30	0.41	0.47	(-0.56, 1.37)	0.395	0.46
Attachment	27	0.59	0.77	(-1.01, 2.19)	0.454	0.01	30	-0.83	0.87	(-2.63, 0.97)	0.353	0.46
Health	27	1.51	0.44	(0.59, 2.42)	0.002	0.40	30	0.41	0.61	(-0.84, 1.66)	0.504	0.45
Role restriction	27	0.29	0.34	(-0.42, 0.99)	0.407	0.13	30	0.72	0.32	(0.05, 1.39)	0.035	0.53
Depression	27	0.51	0.30	(-0.12, 1.14)	0.107	0.20	30	0.14	0.49	(-0.88, 1.16)	0.775	0.44
Spouse	27	0.46	0.34	(-0.25, 1.17)	0.193	0.17	30	0.26	0.38	(-0.52, 1.04)	0.496	0.45
Parent domain	27	0.11	0.06	(-0.02, 0.23)	0.089	0.21	30	0.08	0.09	(-0.10, 0.26)	0.376	0.46
Total stress	27	0.07	0.04	(-0.02, 0.15)	0.115	0.20	30	0.04	0.05	(-0.07, 0.15)	0.467	0.45
Life stress	27	0.02	0.20	(-0.39, 0.43)	0.932	0.10	30	-0.26	0.21	(-0.68, 0.17)	0.230	0.47

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Mental Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6h

Multivariable[†] regression models for MDI[+] at 12 months on PSI at 12 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI [‡]**	**P**	**R2**	**N**	**β**	**SE**	**95% CI [‡]**	**P**	**R2**
Distractibility	43	-0.26	0.36	(-0.99, 0.46)	0.467	0.10	59	0.16	0.36	(-0.57, 0.88)	0.666	0.22
Adaptability	43	-0.04	0.25	(-0.56, 0.47)	0.869	0.09	59	-0.05	0.34	(-0.72, 0.63)	0.894	0.22
Reinforces parents	43	-0.62	0.58	(-1.81, 0.56)	0.293	0.12	59	-1.19	0.71	(-2.61, 0.24)	0.100	0.26
Demandingness	43	-0.20	0.33	(-0.87, 0.48)	0.561	0.10	59	-0.34	0.36	(-1.05, 0.37)	0.342	0.23
Mood	43	-0.58	0.47	(-1.53, 0.36)	0.220	0.13	59	-0.78	0.71	(-2.20, 0.64)	0.278	0.23
Acceptability	43	-0.29	0.48	(-1.25, 0.67)	0.546	0.13	59	-0.96	0.43	(-1.81, -0.10)	0.029	0.28
Child domain	43	-0.06	0.08	(-0.22, 0.10)	0.423	0.11	59	-0.11	0.10	(-0.31, 0.09)	0.286	0.23
Competence	43	0.12	0.22	(-0.31, 0.56)	0.570	0.10	59	-0.47	0.24	(-0.95, -0.00)	0.048	0.27
Isolation	43	0.17	0.33	(-0. 50, 0.83)	0.613	0.10	59	-0.87	0.35	(-1.57, -0.18)	0.015	0.30
Attachment	43	-0.45	0.46	(-1.39, 0.49)	0.334	0.11	59	-0.69	0.62	(-1.93, 0.54)	0.266	0.24
Health	43	0.49	0.45	(-0.43, 1.41)	0.290	0.12	59	-0.24	0.48	(-1.20, 0.72)	0.624	0.22
Role restriction	43	-0.07	0.30	(-0.67, 0.54)	0.823	0.09	59	-0.76	0.26	(-1.28, -0.24)	0.005	0.32
Depression	43	0.02	0.25	(-0.50, 0.53)	0.943	0.09	59	-0.44	0.25	(-0.95, 0.07)	0.091	0.26
Spouse	43	-0.07	0.30	(-0.68, 0.59)	0.820	0.09	59	-0.41	0.31	(-1.02, 0.21)	0.194	0.24
Parent domain	43	0.01	0.06	(-0.11, 0.13)	0.854	0.09	59	-0.13	0.06	(-0.25, -0.02)	0.024	0.29
Total stress	43	-0.01	0.04	(-0.08, 0.06)	0.809	0.09	59	-0.08	0.04	(-0.16, -0.00)	0.046	0.27
Life stress	43	-0.21	0.21	(-0.63, 0.22)	0.331	0.11	59	-0.19	0.19	(-0.57, 0.20)	0.331	0.23

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Mental Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6i

Multivariable[†] regression models for PDI[+] at 12 months on PSI at 3 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	39	0.65	0.52	(-0.40, 1.71)	0.217	0.05	50	0.20	0.47	(-0.74, 1.14)	0.673	0.20
Adaptability	39	-0.03	0.48	(-1.01, 0.95)	0.949	0.00	50	0.33	0.38	(-0.43, 1.09)	0.390	0.21
Reinforces parents	39	-1.13	1.12	(-3.40, 1.15)	0.322	0.03	49	1.31	0.79	(-0.28, 2.90)	0.105	0.27
Demandingness	39	-0.04	0.65	(-1.35, 1.28)	0.955	0.00	50	0.28	0.42	(-0.57, 1.13)	0.511	0.21
Mood	39	-1.23	0.92	(-3.10, 0.63)	0.188	0.05	50	-0.11	0.76	(-1.64, 1.42)	0.885	0.20
Acceptability	39	-0.09	0.91	(-1.95, 1.76)	0.918	0.00	50	0.65	0.62	(-0.60, 1.89)	0.301	0.22
Competence	39	0.15	0.43	(-0.72, 1.02)	0.728	0.01	50	0.17	0.41	(-0.66, 1.00)	0.687	0.20
Isolation	37	-0.06	0.59	(-1.26, 1.13)	0.916	0.02	50	0.09	0.66	(-1.24, 1.41)	0.895	0.20
Attachment	37	-0.90	0.93	(-2.80, 1.00)	0.341	0.04	50	-0.65	0.81	(-2.29, 0.98)	0.424	0.21
Health	37	0.68	0.83	(-1.00, 2.37)	0.417	0.04	50	-0.64	0.72	(-2.09, 0.82)	0.383	0.21
Role restriction	37	-0.26	0.50	(-1.27, 0.74)	0.597	0.02	50	-1.31	0.42	(-2.16, -0.46)	0.003	0.34
Depression	37	-0.06	0.49	(-1.06, 0.94)	0.908	0.02	50	-0.23	0.56	(-1.35, 0.89)	0.683	0.20
Spouse	37	0.05	0.49	(-0.95, 1.06)	0.917	0.02	50	-0.52	0.61	(-1.75, 0.71)	0.400	0.21
Parent domain	37	0.00	0.11	(-0.22, 0.23)	0.969	0.012	50	-0.14	0.12	(-0.39, 0.11)	0.255	0.22
Total stress	37	-0.00	0.07	(-0.15, 0.14)	0.966	0.02	50	-0.00	0.07	(-0.14, 0.13)	0.985	0.20
Life stress	38	0.00	0.24	(-0.49, 0.49)	0.999	0.00	50	-0.51	0.28	(-1.07, 0.06)	0.078	0.25

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Psychomotor Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6j

Multivariable[†] regression models for PDI[+] at 12 months on PSI at 6 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	33	-0.63	0.76	(-2.17, 0.92)	0.415	0.04	44	0.95	0.56	(-0.18, 2.08)	0.098	0.26
Adaptability	33	-0.28	0.55	(-1.41, 0.85)	0.610	0.03	44	0.09	0.68	(-1.29, 1.48)	0.891	0.21
Reinforces parents	33	-2.15	1.29	(-4.80, 0.50)	0.107	0.10	44	2.55	1.34	(-0.15, 5.25)	0.064	0.28
Demandingness	33	-0.08	0.69	(-1.48, 1.33)	0.913	0.02	44	0.02	0.545	(-1.09, 1.12)	0.974	0.21
Mood	33	-1.08	1.10	(-3.34, 1.18	0.335	0.05	44	2.65	0.98	(0.68, 4.63)	0.010	0.34
Acceptability	33	-0.57	1.18	(-2.98, 1.84)	0.633	0.02	44	0.24	0.74	(-1.26, 1.75)	0.746	0.21
Child domain	33	-0.15	0.18	(-0.52, 0.21)	0.399	0.04	44	0.23	0.18	(-0.12, 0.58)	0.198	0.24
Competence	33	0.36	0.51	(-0.68, 1.40)	0.485	0.03	44	-0.24	0.44	(-1.13, 0.65)	0.585	0.20
Isolation	33	0.39	0.80	(-1.25, 2.02)	0.633	0.02	44	-0.56	0.64	(-1.86, 0.74)	0.39	0.23
Attachment	33	-0.69	1.10	(-2.93, 1.56)	0.537	0.03	44	1.35	0.85	(-0.36, 3.07)	0.119	0.26
Health	33	1.08	0.92	(-0.79, 2.96)	0.247	0.062	44	-0.12	0.80	(-1.73, 1.50)	0.884	0.21
Role restriction	33	-0.31	0.56	(-1.45, 0.84)	0.588	0.027	44	-1.19	0.56	(-2.33, -0.06)	0.040	0.29
Depression	33	0.18	0.55	(-0.93, 1.30)	0.739	0.02	44	0.55	0.55	(-0.56, 1.66)	0.323	0.23
Spouse	33	-0.55	0.53	(-1.63, 0.53)	0.304	0.05	44	0.17	0.56	(-0.96, 1.31)	0.759	0.21
Parent domain	33	0.00	0.12	(-0.23, 0.24)	0.966	0.08	44	-0.03	0.13	(-0.30, 0.23)	0.792	0.21
Total stress	33	-0.02	0.08	(-0.18, 0.13)	0.748	0.02	44	0.04	0.08	(-0.13, 0.21)	0.665	0.21
Life stress	33	-0.11	0.27	(-0.71, 0.50)	0.724	0.02	44	-0.66	0.31	(-1.28, -0.03)	0.039	0.29

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Psychomotor Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6k

Multivariable[†] regression models for PDI[+] at 12 months on PSI at 9 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	27	0.90	0.85	(-0.85, 2.65)	0.300	0.06	30	1.11	0.58	(-0.09, 2.31)	0.069	0.38
Adaptability	27	0.83	0.63	(-0.48, 2.13)	0.202	0.08	30	0.76	0.55	(-0.36, 1.89)	0.176	0.34
Reinforces parents	27	2.76	1.40	(-0.13, 5.65)	0.060	0.16	30	1.58	1.045	(-0.58, 3.73)	0.144	0.35
Demandingness	27	0.31	0.73	(-1.19, 1.81)	0.675	0.02	30	-0.78	0.60	(-2.02, 0.46)	0.205	0.34
Mood	27	0.64	1.40	(-2.26, 3.54)	0.652	0.02	30	0.78	1.07	(-1.43, 2.99)	0.475	0.31
Acceptability	27	1.08	1.04	(-1.07, 3.23)	0.308	0.06	30	-0.66	0.82	(-2.34, 1.03)	0.430	0.31
Child domain	27	0.25	0.18	(-0.13, 0.63)	0.187	0.09	30	0.14	0.18	(-0.23, 0.51)	0.444	0.31
Competence	27	0.78	0.45	(-0.16, 1.72)	0.099	0.13	30	-0.02	0.55	(-1.15, 1.11)	0.976	0.29
Isolation	27	0.79	0.72	(-0.69, 2.28)	0.280	0.06	30	0.20	0.74	(-1.32, 1.73)	0.784	0.29
Attachment	27	2.08	1.30	(-0.62, 4.78)	0.124	0.11	30	2.51	1.29	(-0.14, 5.17)	0.063	0.39
Health	27	1.89	0.87	(0.09, 3.68)	0.040	0.18	30	1.59	0.90	(-0.26, 3.45)	0.089	0.37
Role restriction	27	0.79	0.59	(-0.42, 2.00)	0.192	0.09	30	0.40	0.55	(-0.73, 1.53)	0.471	0.31
Depression	27	0.85	0.54	(-0.26, 1.96)	0.126	0.11	30	1.34	0.72	(-0.14, 2.83)	0.075	0.38
Spouse	27	0.31	0.62	(-0.98, 1.59)	0.629	0.03	30	-0.08	0.59	(-1.30, 1.14)	0.895	0.29
Parent domain	27	0.18	0.11	(-0.04, 0.40)	0.111	0.12	30	0.13	0.14	(-0.15, 0.41)	0.348	0.31
Total stress	27	0.12	0.07	(-0.03, 0.27)	0.113	0.12	30	0.08	0.08	(-0.09, 0.25)	0.350	0.32
Life stress	27	0.31	0.34	(-0.40, 1.02)	0.372	0.05	30	-0.71	0.30	(-1.33, -0.09)	0.026	0.42

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Psychomotor Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 6l

Multivariable[†] regression models for PDI[+] at 12 months on PSI at 12 months.

	Healthy infants						CHD infants					
PSI subscale[*]	**N**	**β**	**SE[**]**	**95% CI[‡]**	**P**	**R^2**	**N**	**β**	**SE**	**95% CI[‡]**	**P**	**R^2**
Distractibility	43	.20	.62	(-1.06, 1.46)	0.749	0.01	59	1.25	0.47	(0.31, 2.19)	0.010	0.27
Adaptability	43	0.01	0.44	(-0.88, 0.90)	0.976	0.01	59	0.26	0.47	(-0.67, 1.20)	0.576	0.18
Reinforces parents	43	-0.74	1.02	(-2.80, 1.33)	0.475	0.02	59	0.95	1.00	(-1.05, 2.96)	0.343	0.19
Demandingness	43	-0.33	0.58	(-1.50, 0.83)	0.565	0.02	59	-0.47	0.49	(-1.45, 0.52)	0.345	0.19
Mood	43	-1.22	0.81	(-2.85, 0.41)	0.138	0.07	59	1.96	0.95	(0.05, 3.87)	0.044	0.23
Acceptability	43	-0.91	0.82	(-2.56, 0.75)	0.275	0.04	59	-0.54	0.61	(-1.77, 0.68)	0.378	0.18
Child domain	43	-0.08	0.14	(-0.36, 0.20)	0.562	0.02	59	0.12	0.14	(-0.17, 0.40)	0.412	0.18
Competence	43	0.24	0.37	(-0.51, 1.00)	0.521	0.02	59	-0.38	0.33	(-1.05, 0.29)	0.261	0.19
Isolation	43	0.35	0.57	(-0.80, 1.50)	0.542	0.02	59	-0.86	0.49	(-1.85, 0.13)	0.087	0.22
Attachment	43	0.37	0.81	(-1.27, 2.01)	0.651	0.02	59	0.615	0.86	(-1.10, 2.34)	0.476	0.18
Health	43	1.14	0.78	(-0.43, 2.72)	0.149	0.06	59	-0.42	0.66	(-1.75, 0.90)	0.525	0.18
Role restriction	43	-0.19	0.52	(-1.23, 0.85)	0.714	0.01	59	-1.04	0.36	(-1.76, -0.32)	0.006	0.28
Depression	43	0.24	0.44	(-0.64, 1.13)	0.581	0.02	59	-0.16	0.36	(-0.89, 0.56)	0.652	0.18
Spouse	43	-0.45	0.52	(-1.49, 0.60)	0.391	0.03	59	-0.29	0.43	(-1.16, 0.57)	0.503	0.18
Parent domain	43	0.04	0.10	(-0.16, 0.25)	0.684	0.01	59	-0.11	0.08	(-0.27, 0.05)	0.174	0.20
Total stress	43	-0.00	0.06	(-0.12, 0.12)	0.992	0.01	59	-0.03	0.06	(-0.15, 0.08)	0.544	0.18
Life stress	43	-0.58	0.36	(-1.31, 0.14)	0.109	0.07	59	-0.45	0.26	(-0.97, 0.07)	0.088	0.22

Note. [†]All models are adjusted for infant length Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; [*] Parenting Stress Index subscales scores as the predictors of interest, each represents a separate multivariate model within each group, CHD and healthy; [+]Bayley Psychomotor Development Index scores; [**] Standard Error; [‡]95% Confidence intervals.

Table 7a

Power calculation for Minimal Detectable Differences (MDD) for aim 1[ŧ]

PSI Subscale	**Comparison group**	**Sample size**	**Mean**	**Standard Deviation**	**MDD**
Visit 2 (three months of age)					
Reinforces parents	Healthy	63	7.75	2.09	1.2
	CHD	65	8.08	2.72	
Acceptability	Healthy	63	10.44	2.70	1.7
	CHD	66	11.32	4.13	
Child domain	Healthy	63	88.51	16.49	9.5
	CHD	66	95.52	21.37	
Competence	Healthy	63	22.10	5.75	2.8
	CHD	66	24.17	5.54	
Spouse	Healthy	60	15.62	4.50	2.4
	CHD	66	16.77	5.04	
Parent domain	Healthy	60	107.23	21.57	10.7
	CHD	66	112.42	20.76	
Total stress	Healthy	60	195.85	34.17	18.3
	CHD	66	207.94	38.04	
Visit 3 (six months of age)					
Demandingness	Healthy	46	15.07	4.02	2.4
	CHD	54	17.52	4.58	
Acceptability	Healthy	46	10.20	2.46	1.7
	CHD	54	11.50	3.48	
Child domain	Healthy	46	88.22	16.73	9.4
	CHD	54	91.67	16.24	
Health	Healthy	46	11.96	3.03	1.7
	CHD	54	12.00	3.02	
Depression	Healthy	46	17.02	4.95	2.7
	CHD	54	16.63	4.46	
Parent domain	Healthy	46	108.80	23.48	12.6
	CHD	54	109.87	20.59	
Total stress	Healthy	46	197.02	36.87	19.8
	CHD	54	201.54	32.13	
Visit 4 (nine months of age)					
Adaptability	Healthy	39	22.85	4.56	2.8
	CHD	56	23.27	4.90	
Demandingness	Healthy	39	15.26	4.15	2.5
	CHD	56	18.16	4.25	

Child domain	Healthy	39	89.95	16.08	9.6
	CHD	56	93.50	16.47	
Isolation	Healthy	39	11.82	4.05	2.4
	CHD	56	12.20	3.78	
Role restriction	Healthy	39	18.62	4.68	2.8
	CHD	56	17.48	4.91	
Parent domain	Healthy	39	110.08	24.33	13.4
	CHD	56	109.23	20.69	
Total stress	Healthy	39	200.03	37.81	21.6
	CHD	56	202.73	34.44	
Life stress	Healthy	39	7.08	8.51	5
	CHD	56	10.32	8.32	
Visit 5 (twelve months of age)					
Demandingness	Healthy	56	15.38	4.08	2.2
	CHD	65	17.88	4.43	
Mood	Healthy	56	9.04	2.69	3.3
	CHD	65	8.86	2.33	
Child domain	Healthy	56	92.14	16.17	8.2
	CHD	65	93.92	15.71	
Attachment	Healthy	55	10.78	2.80	1.4
	CHD	65	9.89	2.43	
Role restriction	Healthy	55	17.64	4.67	2.6
	CHD	65	16.71	5.53	
Parent domain	Healthy	55	107.75	22.34	12.3
	CHD	65	107.02	25.68	
Total stress	Healthy	55	200.11	36.24	19.1
	CHD	65	200.94	37.55	

Note. [†] Computed via two-sample independent t-tests, $\alpha=0.05$, 80% power.

Table 7b

Post-hoc power calculations for Aim 2.*

PSI subscale	N	Mean (β)	SD**	Power	Effect size
Distractibility	124	0.05	3.09	0.05	0.016
Adaptability	124	-0.34	2.81	0.26	0.121
Reinforces parents	124	-0.32	1.56	0.61	0.205
Demandingness	124	-0.38	2.4	0.41	0.158
Mood	124	-0.49	1.72	0.87	0.285
Acceptability	124	-0.14	2.17	0.11	0.065
Child domain	124	-1.55	8.6	0.50	0.180
Competence	124	-0.67	2.77	0.75	0.242
Isolation	123	-0.21	1.85	0.23	0.114
Attachment	123	-0.39	1.69	0.70	0.231
Health	123	-0.18	1.84	0.18	0.098
Role restriction	123	-0.70	2.74	0.79	0.255
Depression	123	-0.34	2.82	0.26	0.121
Spouse	123	-0.06	2.44	0.06	0.025
Parent domain	123	-2.41	9.82	0.76	0.245
Total stress	123	-3.69	14.94	0.76	0.247
Life stress	123	1.48	4.7	0.93	0.315

Note. † Solved for power via two-sided Wilcoxon tests, α=0.05; **Standard Deviation, derived from the residual variance estimates.

Table 7c

Post-hoc power calculation examples for Aim 3.*

Note. *Computed via F-Tests, α=0.05, 80% power; †Models are adjusted for infant length

Regression model†	Healthy N	Healthy R^2 for control variables	Healthy MD R^2 for PSI Subscale	CHD N	CHD R^2 for control variables	CHD MD R^2 for PSI Subscale
MDI+ at 6mo on Total Stress at 3mo	37	0.24	0.14	34	0.37	0.13
MDI at 6mo on Total Stress at 6mo	33	0.32	0.14	41	0.29	0.12
PDI+ at 6mo on Total Stress at 3mo	36	0.14	0.16	35	0.25	0.14
PDI at 6mo on Total Stress at 6mo	32	0.05	0.20	42	0.23	0.13
MDI at 12mo on Total Stress at 3mo	37	0.12	0.16	50	0.23	0.11
MDI at 12mo on Total Stress at 6mo	33	0.25	0.15	44	0.27	0.12
MDI at 12mo on Total Stress at 9mo	27	0.11	0.21	30	0.48	0.11
MDI at 12mo on Total Stress at 12mo	43	0.10	0.14	59	0.22	0.09
PDI at 12mo on Total Stress at 3mo	37	0.05	0.18	50	0.20	0.11
PDI at 12mo on Total Stress at 6mo	33	0.04	0.19	44	0.26	0.12
PDI at 12mo on Total Stress at 9mo	27	0.06	0.23	30	0.38	0.14
PDI at 12mo on Total Stress at 12mo	43	0.01	0.16	59	0.28	0.09

Z-scores at the measured PSI time point, and gestational age in the healthy group; and for infant length Z-scores, gestational age, and length of hospital stay in the CHD group; +Bayley's Mental and Psychomotor Development Index scores. MD= minimal detectable.

Table 8

*Short descriptions of the PSI subscales**

Child Domain subscales	**Short description**
Adaptability	Difficulty adjusting to changes, inflexible
Distractibility	ADHD type behaviors
Demandingness	Demands requiring accommodation or attention
Mood	Moodiness, crying, displays of unhappiness
Acceptability	Behaviors that do not match parent's expectations, hoped-for child
Reinforces parent	Parent does not experience positive reinforcement from interactions with their child
Parent domain subscales	
Competence	Sense of competency in the parental role
Isolation	Lack of social support for their role as parent
Health	Impact of physical health on parenting
Role restriction	Impact of the restrictions parenting places on their choices or freedom
Depression	Impact of depression and feelings of guilt on their parenting behavior
Spouse	Help and emotional support from the other parent

Note. *adapted from Abidin, R. E., Austin, W. G., & Flens, J. R. (2013).

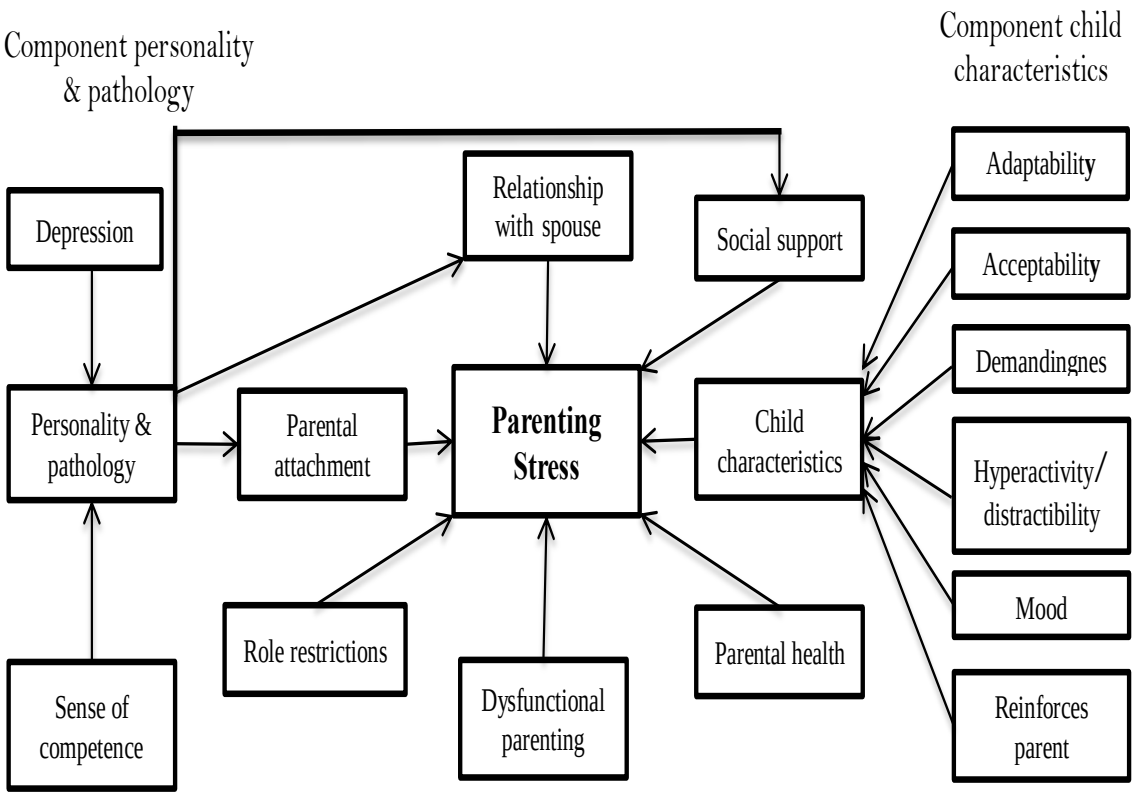

Figure 1: Theoretical model for Parenting Stress (Abidin, 1976)

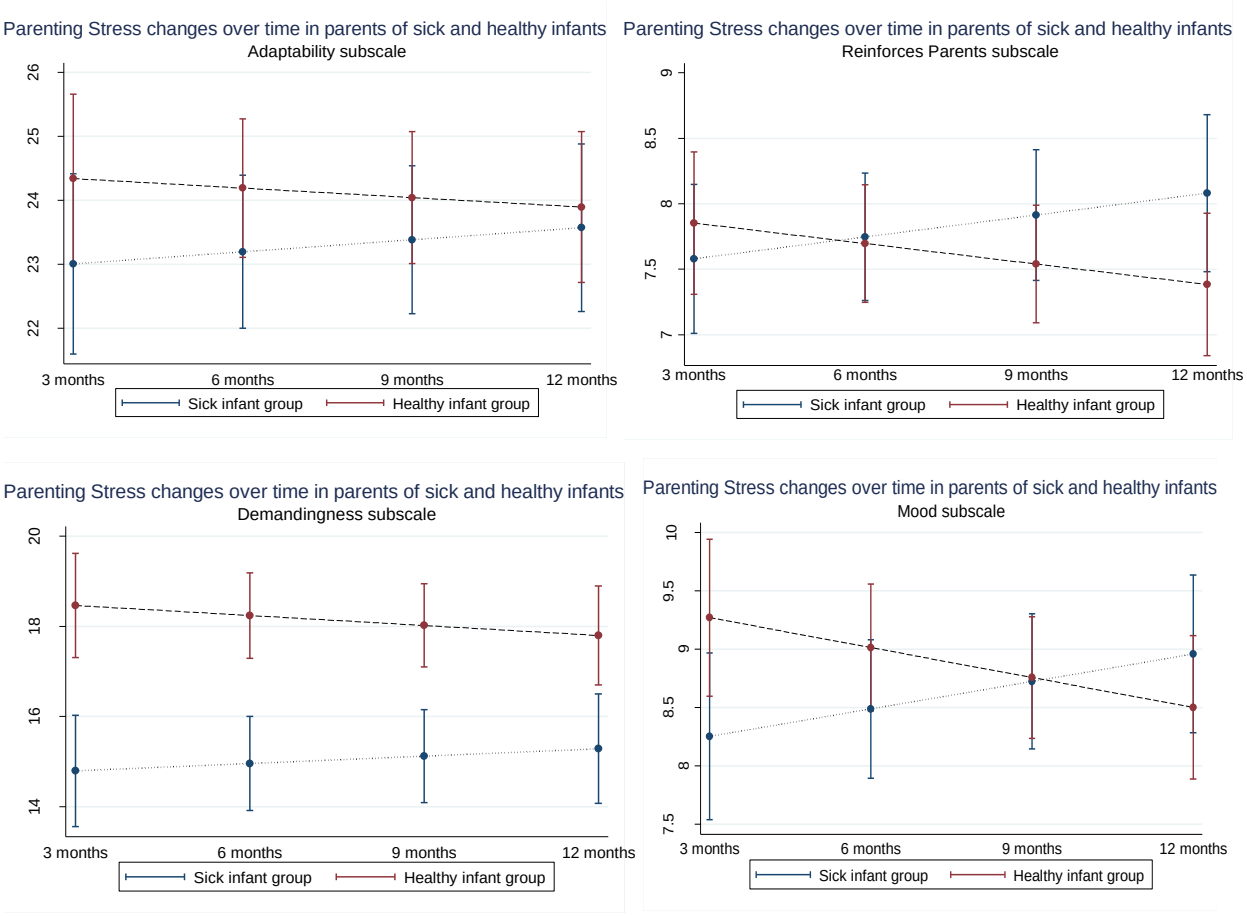

Figure 3. Parenting stress changes over time in parents of CHD and healthy infants.

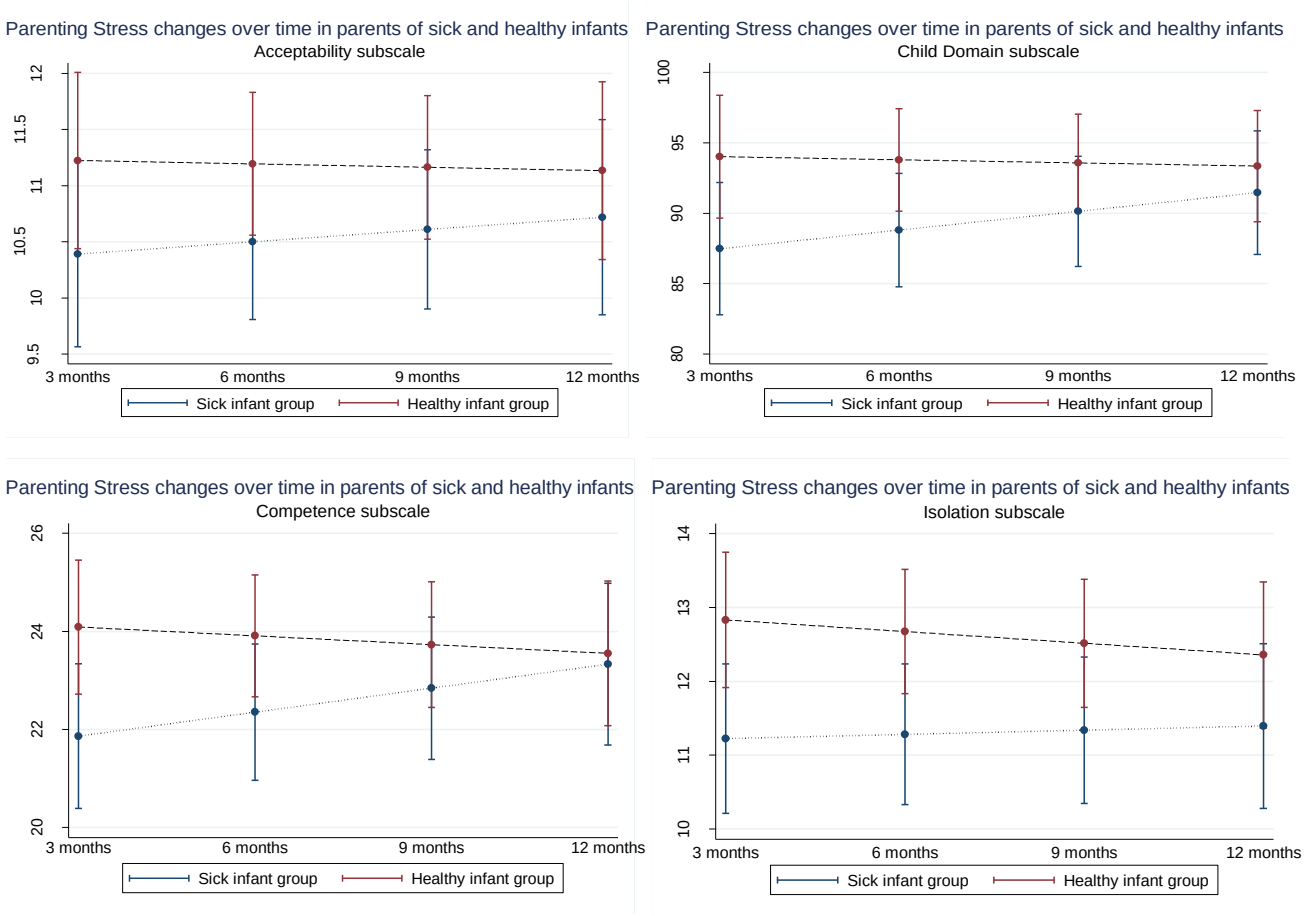

Figure 3. Parenting stress changes over time in parents of CHD and healthy infants.

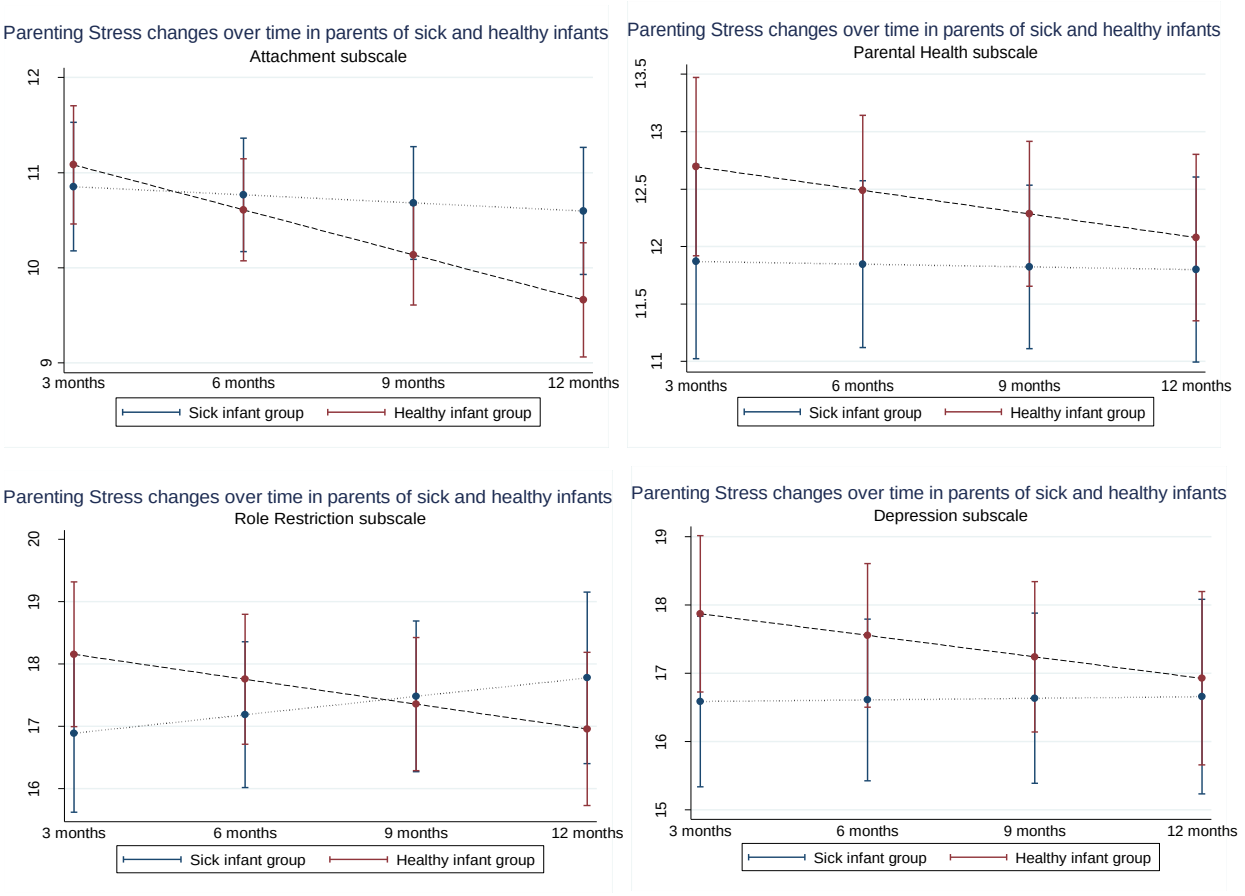

Figure 3. Parenting stress changes over time in parents of CHD and healthy infants.

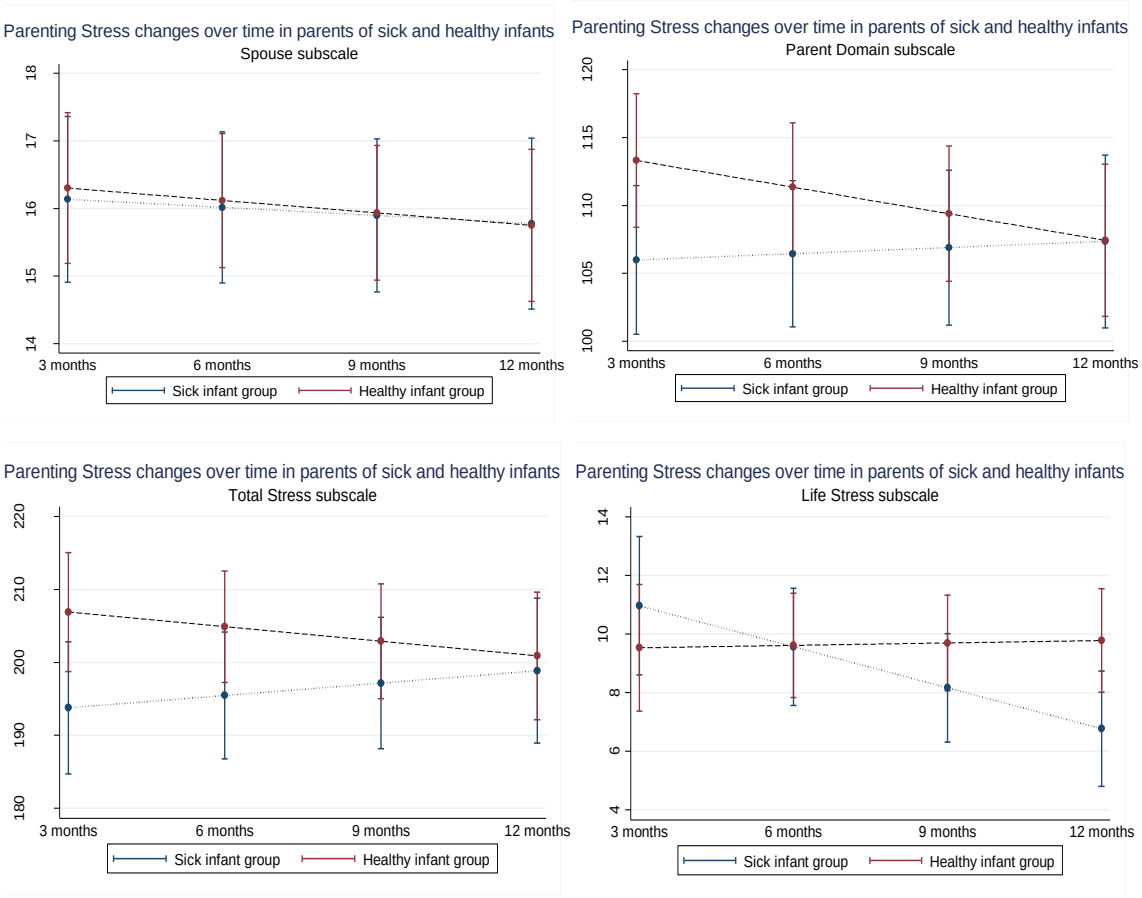

Figure 3. Parenting stress changes over time in parents of CHD and healthy infants.

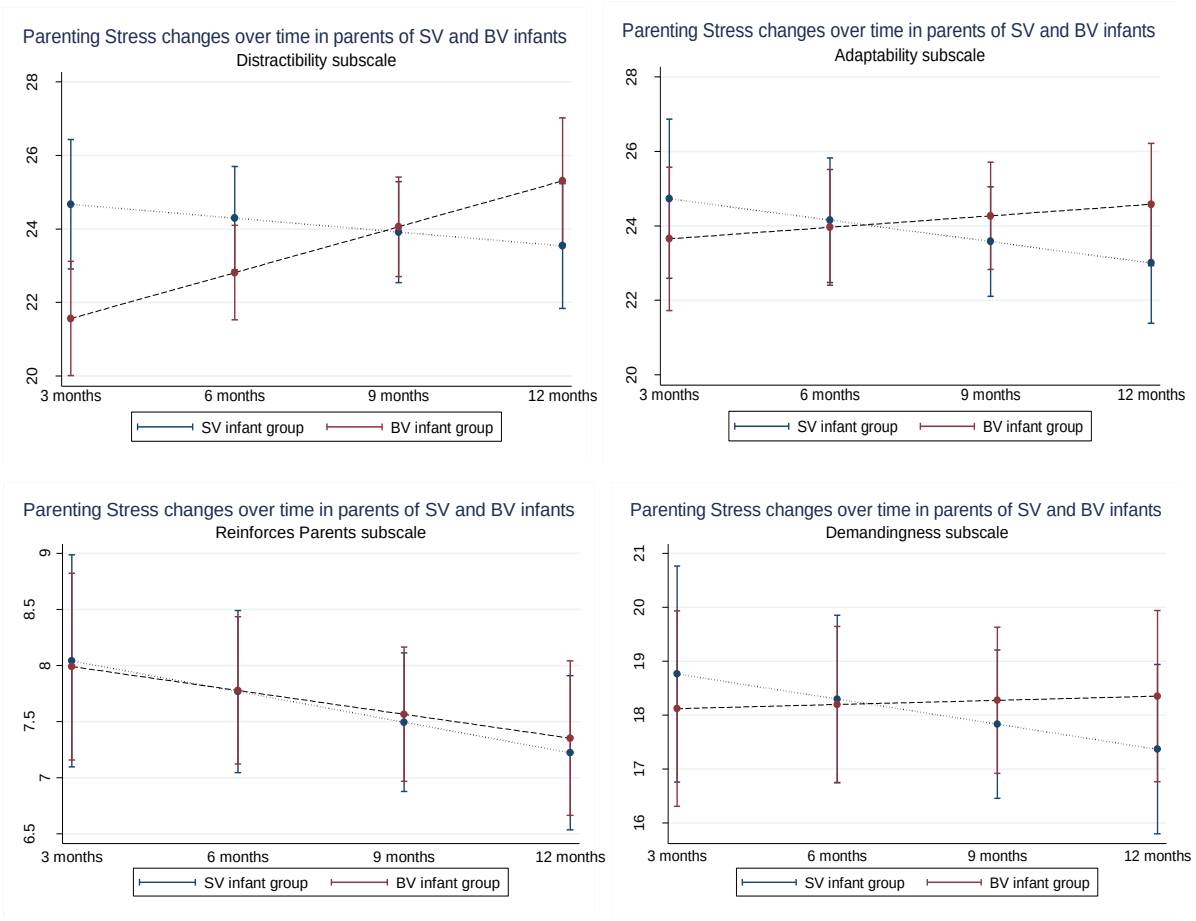

Figure 4. Parenting stress changes over time in parents of SV and BV infants.

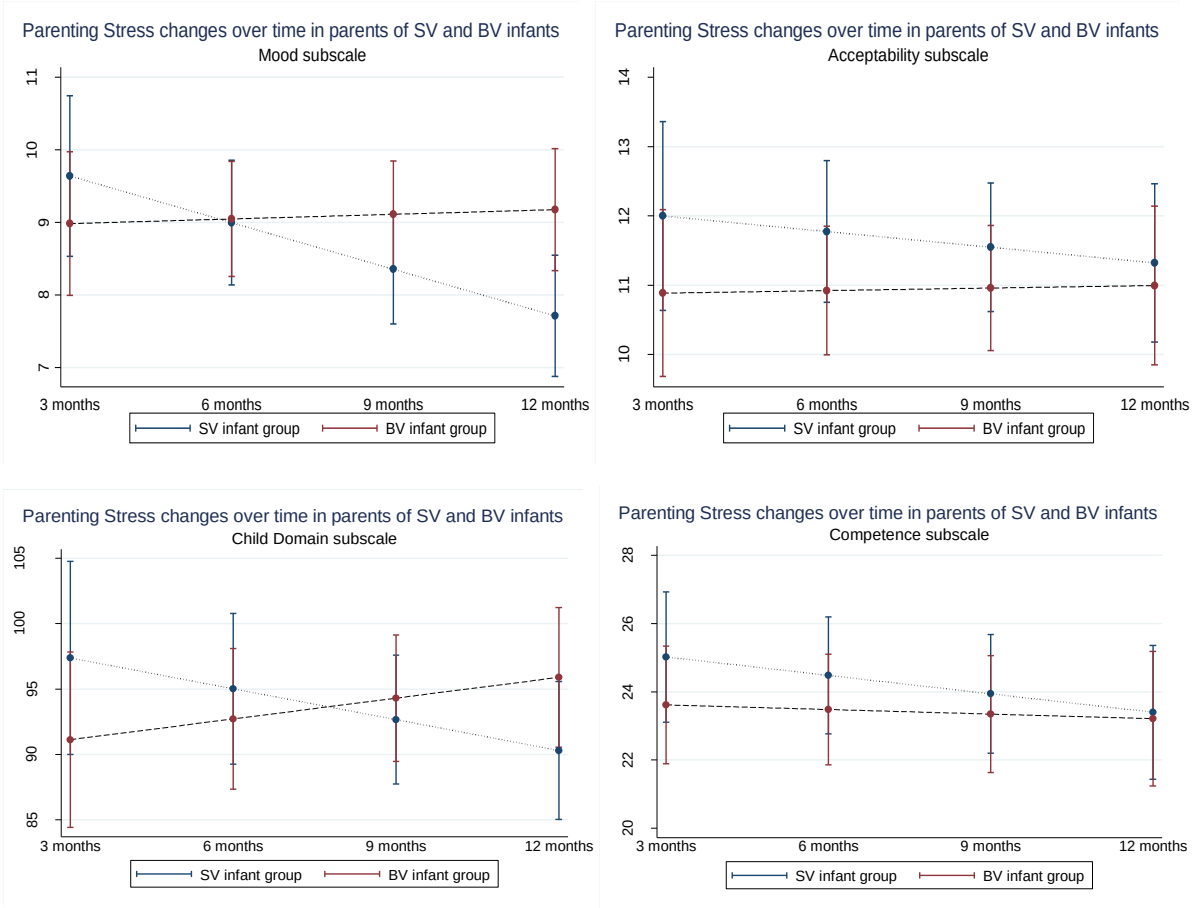

Figure 4. Parenting stress changes over time in parents of SV and BV infants.

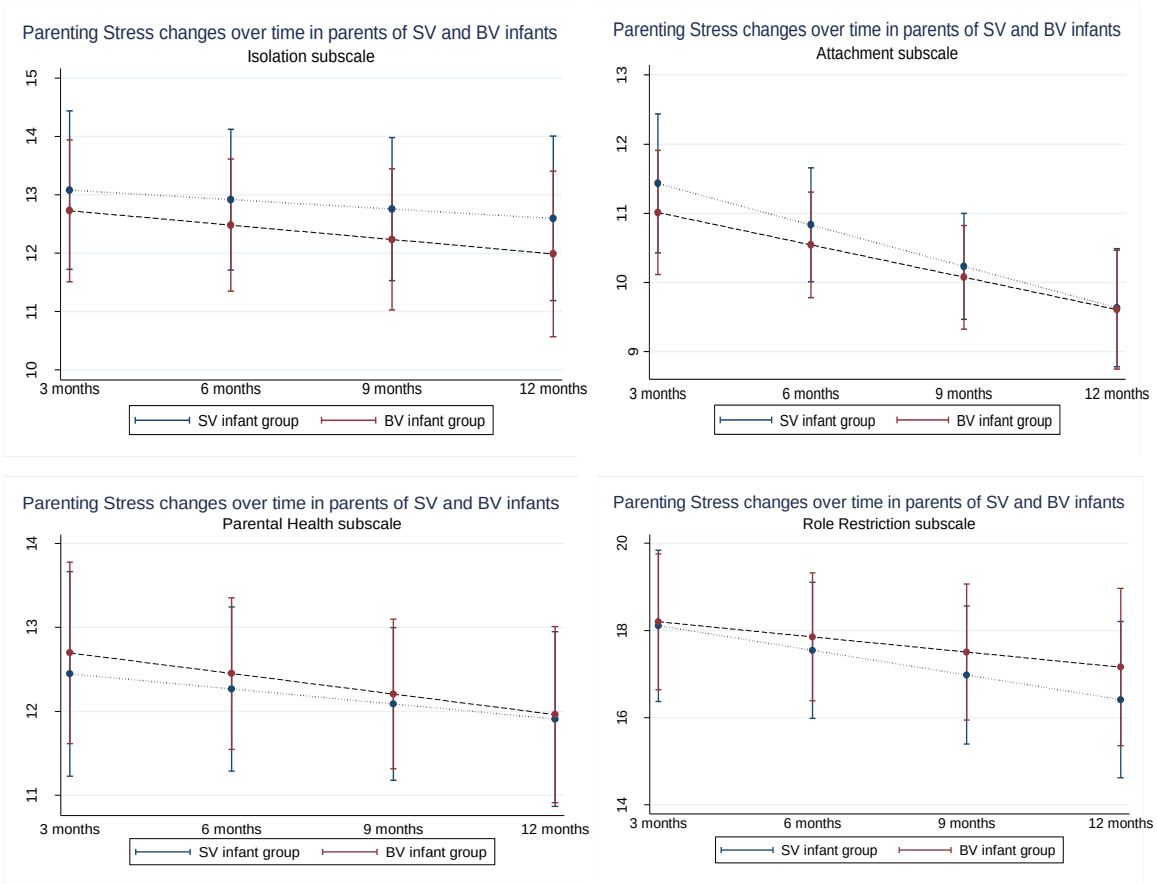

Figure 4. Parenting stress changes over time in parents of SV and BV infants.

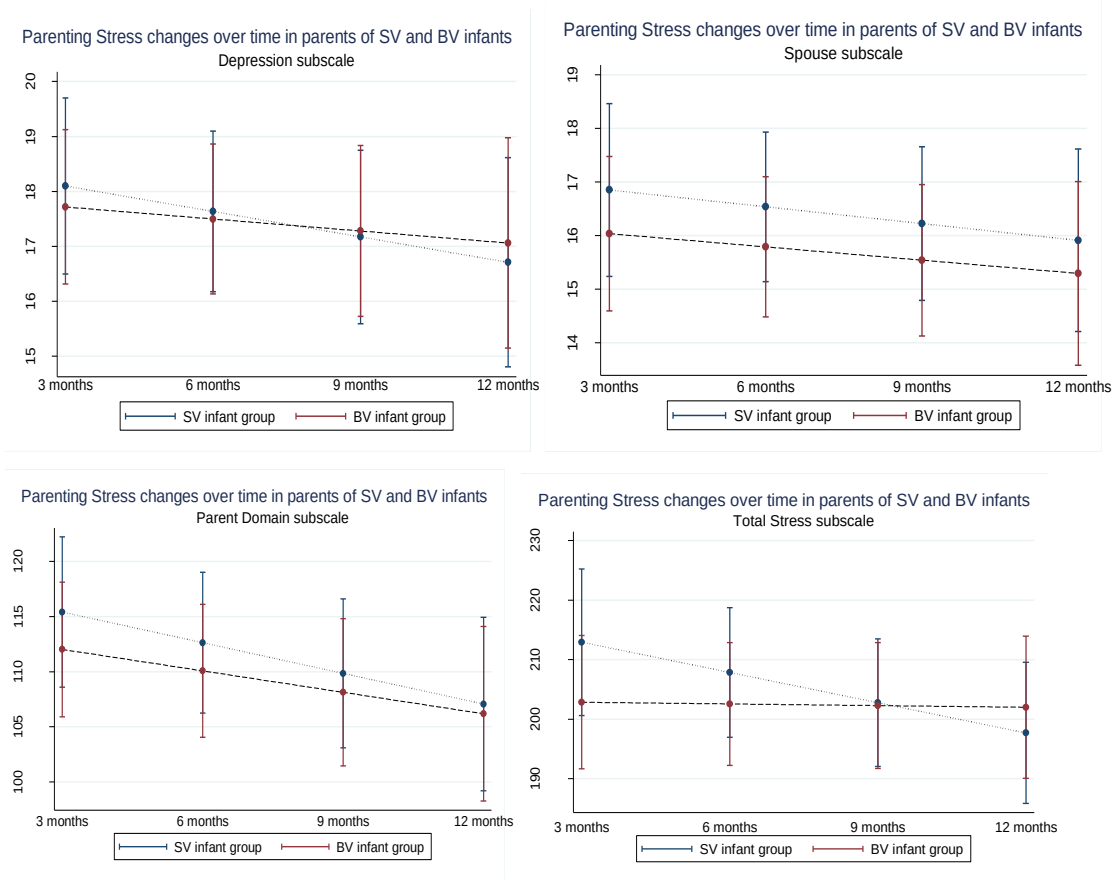

Figure 4. Parenting stress changes over time in parents of SV and BV infants.